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COLLEGE INTERNATIONAL DES CHIRURGIENS

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Original Articles

Nonunion of Supracondylar Fractures of the Femur

CARLO SCUDERI, M D, PH D, F A C S, F I C S AND
ANTHONY IPPOLITO, M D, F A C S, F I C S†
CHICAGO

THE purpose of this paper is to discuss the prevention and treatment of nonunion of supracondylar fractures of the femur, a subject which, according to the literature, has been infrequently discussed. In our opinion, for certain types of supracondylar fractures, early operation with Blade plate fixation is most important in order to obtain adequate reduction and to minimize postoperative immobilization of the knee joint with the resulting fibrosis.

Nonunion of a supracondylar fracture of the femur is not very common, but when it does occur it presents a real problem in surgical treatment. We wish to present a method that has been tried with no failures in a relatively small number of cases and should be of service to other surgeons faced with a similar problem

Historical Background.—Only two references are made in the literature to the treatment of fractures of the distal end of the femur by Blade plate fixation, one by Umanski[1] in April 1948 and one by Dr. Louis Wright[2] in June of the same year. As far as we know, this is the first publication of a suggested method of treatment of nonunion of supracondylar fracture by the use of a Blade plate and massive bone graft according to the technic described in this article.

Pathologic Picture.—Fractures of the supracondylar region of the knee joint are as a rule of a transverse type but do not necessarily follow this pattern. On a number of occasions we have seen fresh fractures of this area with an oblique fracture line, and frequently associated with a dicondylar fracture of either the "T" or the "Y" type. Sometimes there is considerable displacement of one of the condylar fragments. This in itself constitutes a real test of surgical skill for its adequate reapposition.

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†Deceased

Read at the Sixteenth Annual Assembly of the United States Chapter, International College of Surgeons Chicago Sept 10 1951

Submitted for publication Oct 5, 1951

Unless good internal fixation is obtained, the distal fragment usually produces a posterior rotation impossible to overcome, in some cases even during open reduction, because of the difficulty of maintaining immobilization of the distal fragment during the operation. All too commonly the distal end of the proximal fragment protrudes into the quadriceps muscle or the quadriceps tendon and cannot be released from this tissue by any but surgical means. In spite of the fact that most textbooks speak at great length of rupture of the popliteal artery and damage to the anterior and posterior tibial nerves, it has been a rare occurrence in our experience. In only 1 case does the senior author remember damage of the popliteal artery occurring as a direct result of a supracondylar fracture with posterior rotation of the distal fragment. This experience extends over a period of about twenty years.

Nonunion in the supracondylar area is usually a sclerosing type of nonunion. There is an abundance of overgrowth of callus on both the medial and the lateral aspect of the femur, with incomplete solidification across the area of the fracture. As time goes on, the calcium deposit becomes greater, but for some unknown reason true consolidation of the two fractured fragments never occurs. At operation it is not uncommon to find the bone so sclerotic that it is difficult to cut with a chisel, a saw or a drill, and the bone fragments left by the operative intervention have the appearance of chalk, without any gross evidence of vascularity. In several instances this sclerosing type of nonunion has actually obstructed visualization of the nonunion; in 1 particular instance the diagnosis of solid bony union was made, only to be proved erroneous when a few weeks after the patient began weight bearing without protection.

[illegible].—Everyone realizes that a fracture of this magnitude in the vicinity of the knee joint is bound to result in a considerable amount of periarticular fibrosis with limitation of motion, especially in flexion. In patients who have been treated with the knee bent for a long period there is also the additional serious complication of inability of complete extension. This constitutes a real disability because the man not only has to walk poorly but obtains a potential shortening of the leg in walking, which results in an awkward gait, with a serious lurch of the pelvis, and a poor posture. The disability following this type of fracture is long-lasting, extending usually eight to twelve months under the most favorable conditions. One can safely say that the disability certainly lasts longer than that associated with a fracture of the neck of the femur that terminates successfully with solid bony union, or a fracture of the shaft of the femur which is uncomplicated.

Treatment.—It is an axiomatic principle that the sooner the fracture is reduced and placed in anatomic position, the less is the reaction of the tissues to the trauma and the quicker the healing. Therefore it is ideal, circumstances permitting, that supracondylar fractures be reduced anatomically as quickly as possible with sufficient internal fixation, so that early mobilization of the knee joint can be undertaken. By this method the period of convalescence will be shortened and the resulting loss of knee motion minimized. Under the best circumstances one must accept a certain amount of loss of knee motion. Frequently this amounts to 30 to 60 degrees of motion which is never regained.

Postoperatively, whether in a case of fresh fracture or a case of long-standing nonunion, physical therapy should be instituted as soon as there is enough evidence of callus formation to assure one that the fragments will not become displaced. At first it is done passively by the patient with a Pearson attachment; later it is done passively by the physiotherapist, and finally it is done actively

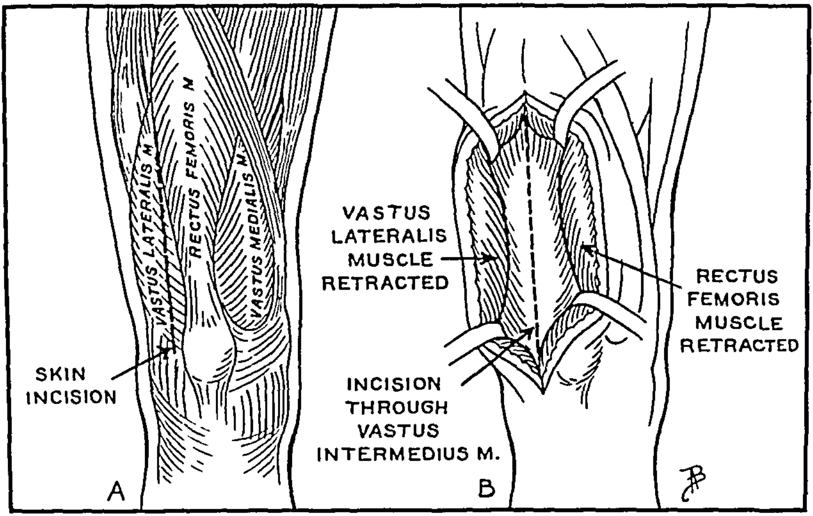
VASTUS LATERALIS M.
RECTUS FEMORIS M.
VASTUS MEDIALIS M.
SKIN INCISION
A
VASTUS LATERALIS MUSCLE RETRACTED
RECTUS FEMORIS MUSCLE RETRACTED
INCISION THROUGH VASTUS INTERMEDIUS M.
B

Fig 1—*A*, rectus femoris muscle separated from vastus medialis through line of cleavage *B*, both muscles retracted and incision through vastus intermedius muscle indicated by dotted line

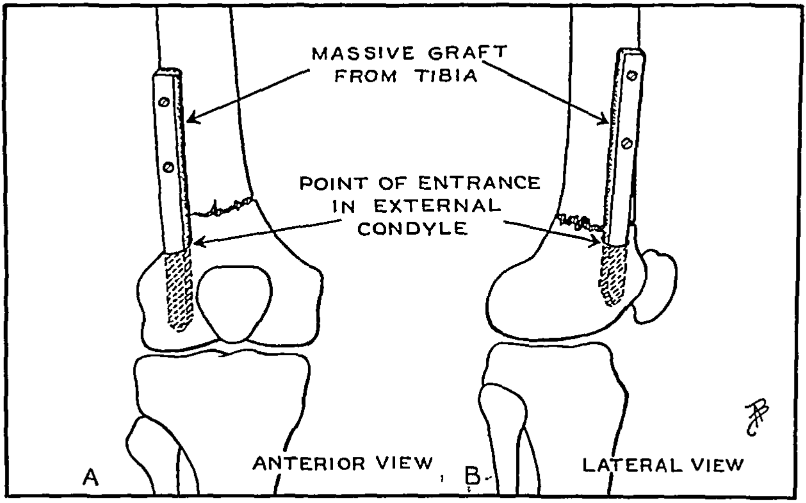

Fig 2—Anterior (*A*) and lateral (*B*) views of the insertion of bone graft

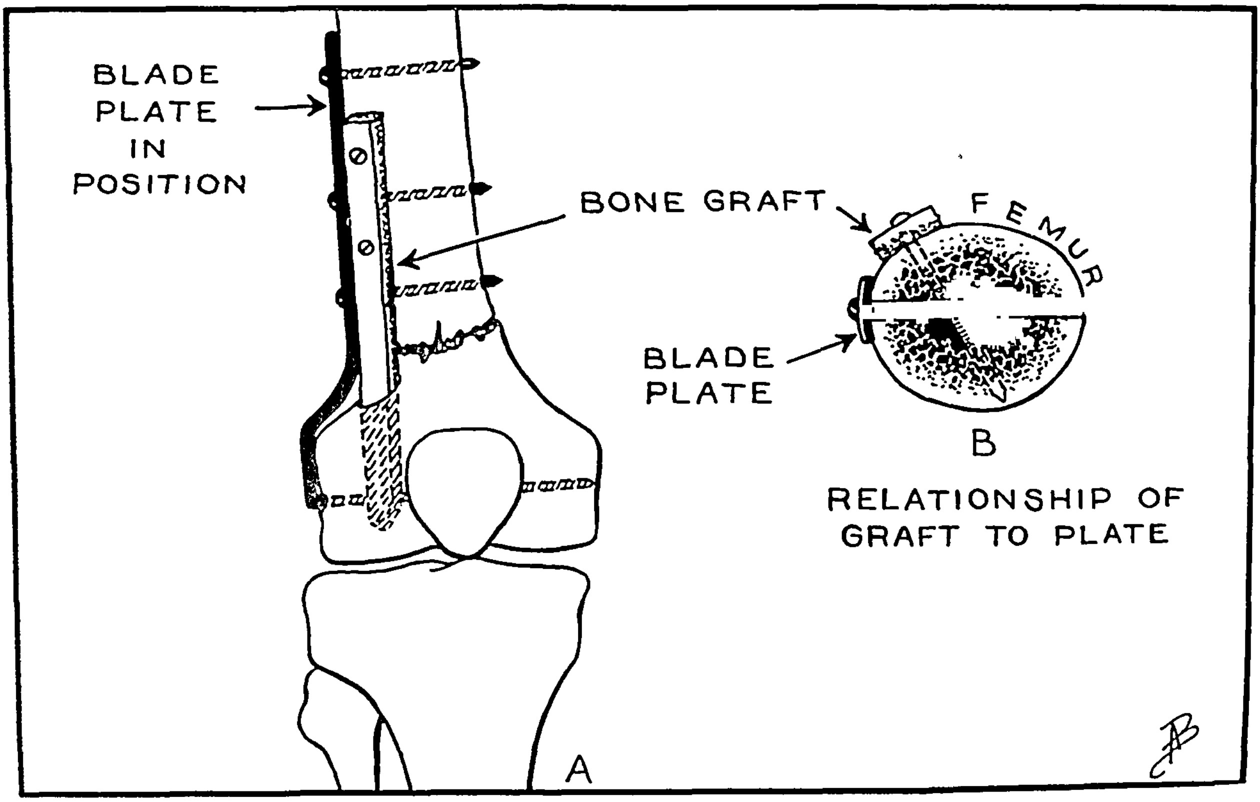

Fig. 3.—*A*, application of Blade plate to permit early mobiliation of knee joint and insure against fracture of graft. *B*, diagram showing relation of graft to Blade plate.

by the patient. This requires a great deal of cooperation between the patient and the physiotherapist, and only by intelligent cooperation and a thorough understanding of the problem can one hope to attain the optimal end result.

The operative procedure through the soft tissues is identical in "fresh" and "old" cases. It has been our experience that the best method of approaching this area is through an anterolateral incision about 6 to 8 inches (15.2 to 20.3 cm.) in length, depending on the underlying pathologic condition. The lateral margin of the rectus femoris is carefully isolated and it is separated from the vastus lateralis through the line of cleavage (Fig. 1). This brings one down to the vastus intermedius, which in this area is not very vascular. The fibers are cut and all the [illegible] carefully controlled. We prefer [illegible] type of operation under an [illegible] pneumatic constrictor, so that [illegible]. The operation can then proceed much more speedily, and there is minimal loss of blood, which decreases the possibility of postoperative shock.

The incision may have to be extended down to the lateral margin of the patella. One need not fear entering the knee joint if necessary. Adequate exposure is imperative for successful reduction. A subperiosteal dissection brings the femoral fragments into view, and by retraction all the important structures in the popliteal area are kept away from the operative field. It is necessary to be able to visualize not only the lateral but the anterior aspect of the femur in the area of the fracture. This can be successfully done by drawing the patella and rectus femoris muscle medially. Careful reapposition of the bone is obtained. We have found it advantageous to use the Blade plate in an adequate size so that the blade portion can penetrate through the external condyle into the medial condyle of the femur and give good fixation. The right-angle portion of the Blade plate, with the holes for screw insertion, is then carefully bent to conform with the femur, after which the screws are inserted for firm immobilization of the proximal fragment In a fresh fracture this can be achieved without too much difficulty and without too extensive dissection or curettage of the injured bone.

If there are some defects in the area because of comminution, it has been our experience that osteogenesis is helped con*siderably if one takes a mass of osteo*periosteal grafts from the shaft of the femur and carefully packs them into the defective area. The closure of the muscle layers and fascia is done with No 000 chromic catgut, and black silk is used for skin closure. The wound is covered with sterile dressings, and then a moderate compression bandage is applied. We use fluffed gauze or mechanic's waste with a superimposed elastic bandage of the Ace or Tensor type. This minimizes postoperative oozing and prevents postoperative edema, which occurs if moderate pressure is not exerted. In all cases the member is placed in Buck's extension with 5 to 10 pounds (2.3 to 4.5 Kg.) of weight, or, if the skin has been poor in the area of the calf, a Steinman pin is inserted through either the tibial tuberosity or the os calcis. Postoperative immobilization in traction is necessary until evidence of *callus formation appears, which as a rule* is after about eight weeks. Physical therapy can be instituted as soon as there is callus formation, so that one need not fear loss of the position of the fractures and disruption of the early stages of union.

Usually, at the end of about ten to twelve weeks, the patient has regained a

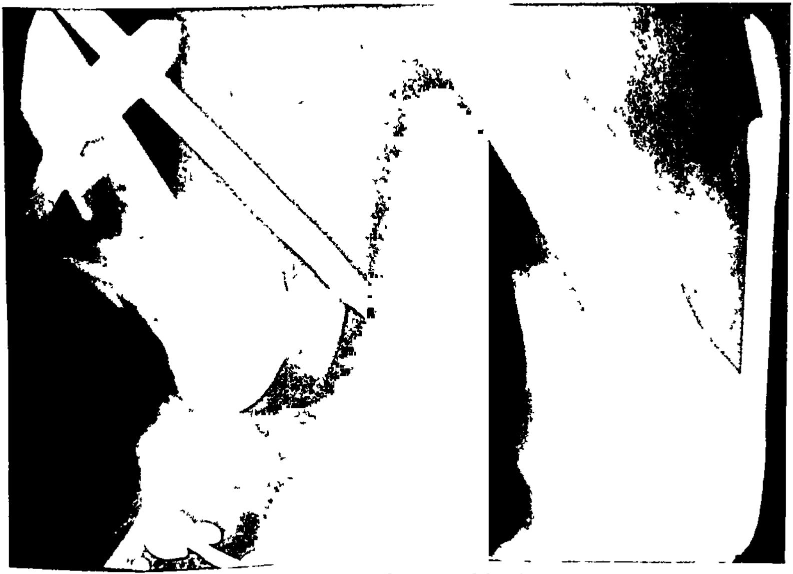

Fig. 4 (J. W., Case 1).—Roentgenograms of supracondylar fracture due to fall (see text).

Fig. 5 (J. W., Case 1).—Roentgenograms taken seven months after treatment.

substantial amount of knee motion with the assistance of the physiotherapist and the use of the Pearson attachment by the patient while he is in bed. An ischial weight-bearing caliper is then used for about six to seven months from the date of operation, until there is no question that solid bony union exists, at which time the brace is completely discarded.

In none of the patients treated by early operation and Blade fixation did we fail to obtain solid bony union.

Nonunions of the supracondylar area present a different problem. The knee joint as a rule is very rigid; the patient is unable to walk, because of instability, and a sclerosing nonunion is usually visible on the roentgenogram and at operation. The bone ends are avascular, the bone is very hard and chalklike in consistency and all evidence of bone regeneration has completely ceased. In these cases it is felt that the patient requires not only adequate immobilization but a substantial massive bone graft to produce solid bony union. The operative approach is through the same layers of tissues and the same type of incision as has been described for fresh fractures (Fig. 1). A massive bone graft is removed from the same tibia, usually 6 to 8 inches (15.2 to 20.3 cm.) in length, depending on the area requiring the graft and the extent of pathologic change. The bone graft is inserted on the anterolateral aspect of the femur and the condyles (Fig. 2). It is necessary that the rounded surface of the femur be flattened so as to take a flat

[illegible] (F. P., Case 2).—A, bilateral fracture of femur treated in Pearson attachment with Buck's [illegible] roentgenograms taken after four and one-half months, showing sufficient callus for freedom of use.

A

B

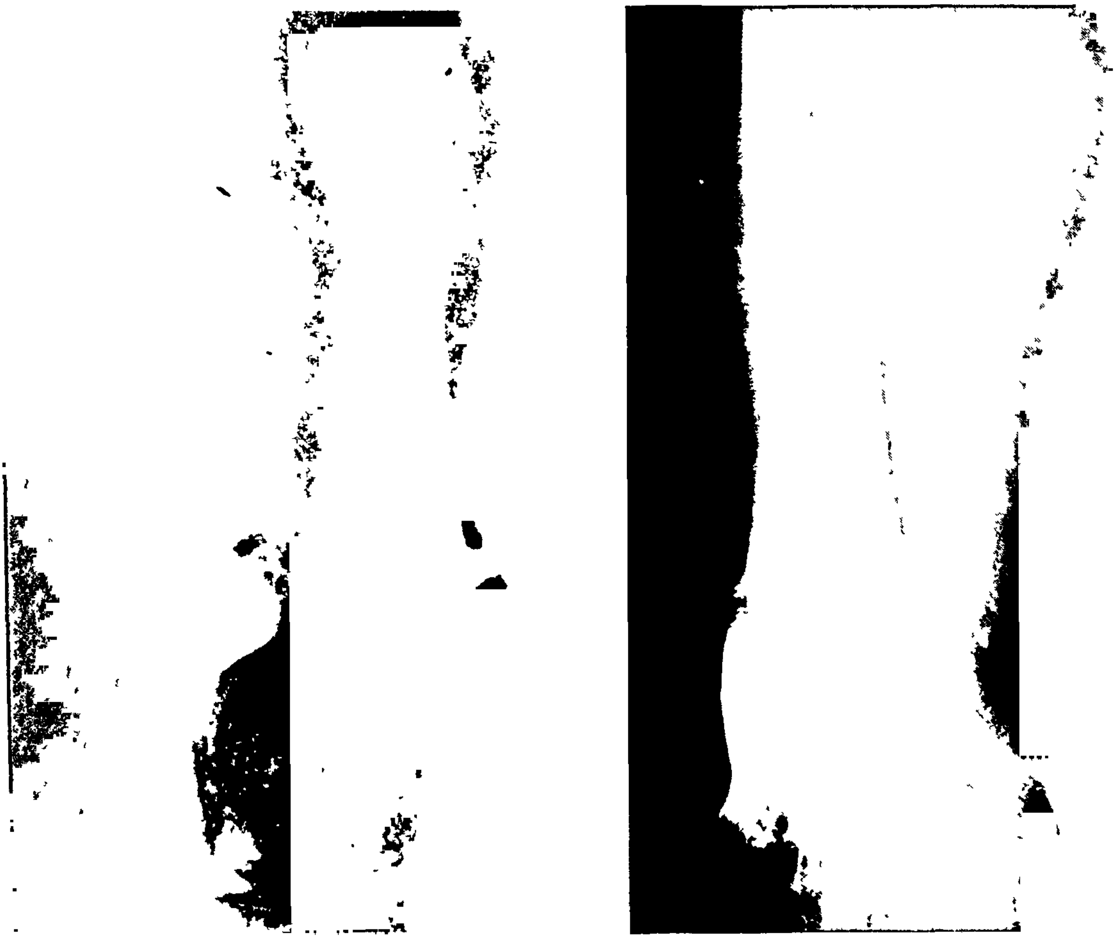

Fig. 7 (W. S., Case 3).—Sclerosing nonunion of fracture unsuccessfully treated by open reduction (see text).

graft and obtain the maximum amount of contact of bone to bone. If one makes a tunnel into the condyles and sharpens the end of the graft, the massive graft can be driven into the femoral condyles and inserted under direct vision until it just reaches the articulation of the femur. The farther distally the graft can be inserted, the better is the fixation of the graft and the better the chances of a good solid union. When the graft is inserted in this slot, fixation of the massive graft is necessary only by screws on the proximal fragment. The distal fragment is completely surrounded by cancellous bone, which heals rapidly, holds the cortical bone graft from the tibia firmly and requires no further fixation. In order to permit early mobilization of the knee joint and to assure against a fracture of the graft, a Blade plate is placed on the lateral aspect of the femur (Fig. 3). This has to be bent to conform with the contours of the femoral condyle and the lateral aspect of the femoral curvature. Fixation is then obtained by the use of two, three or four screws into the handle of the Blade plate.

The closure is the same as for a fresh fracture, as is the treatment. It is most important, however, in the presence of an old nonunion that mobilization be started as early as feasible, because there already exists a considerable amount of periarticular fibrosis and one does not wish to allow more to accumulate. Bedside physical therapy is very important even while the

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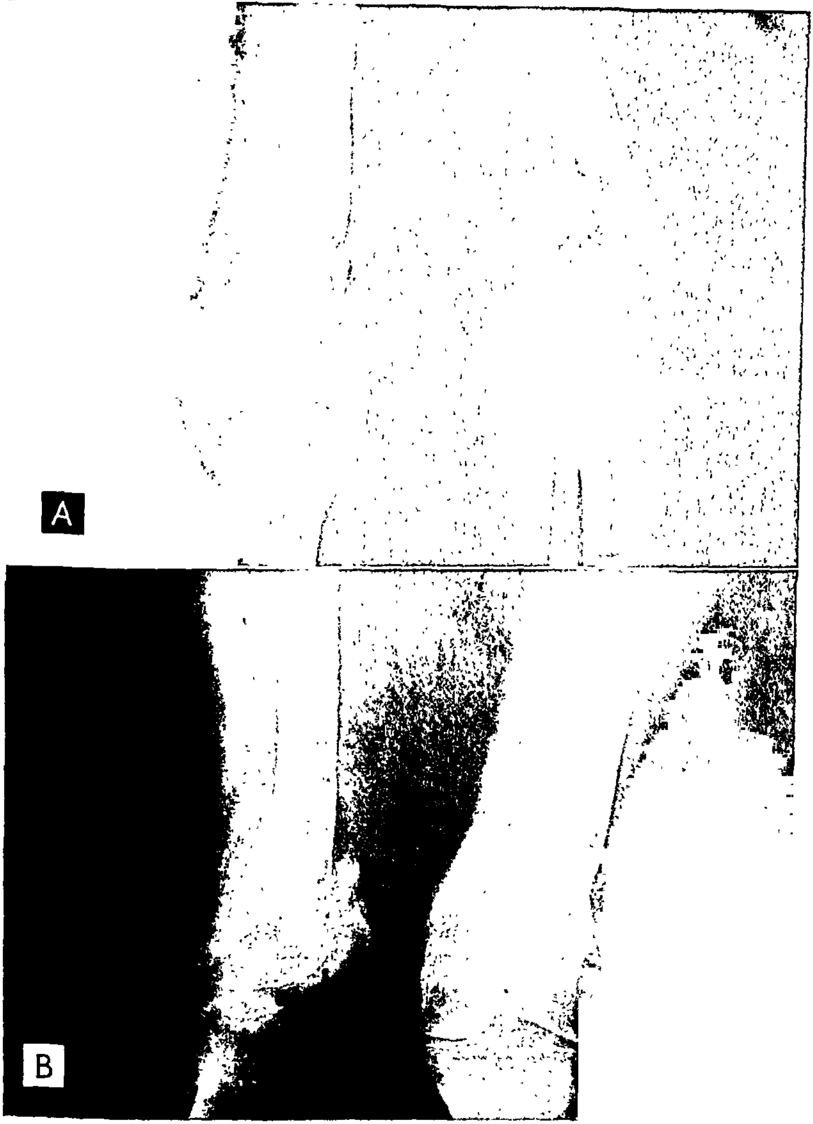
[illegible] F., Case 4).—A, fracture sustained in automobile accident. B, condition nearly two years later.


A
B

patient is in traction, especially therapy in the form of quadriceps setting exercises.

In our cases the usual period from insertion of the bone graft until the patient can walk without the assistance of a brace is seven to nine months.

REPORT OF CASES

CASE 1 (J. W., aged 65).—The patient was injured on Oct. 15, 1950, when he fell down a flight of stairs and sustained a supracondylar fracture with posterior displacement of the distal fragment. This patient was treated with a tibial pin for traction with 15 pounds (6.8 Kg.) of weight. Efforts were made to obtain reduction by use of slings, without success (Fig. 4).

On November 2 the patient was operated on, and a Blade plate was inserted and the patient treated in traction. After about two weeks it was necessary to remove the Steinman pin because of a low grade infection around one side of the pin. The patient was then placed in Buck's extension with 10 pounds (4.5 Kg.) of weight. This was one of the fresh fractures treated by Blade plate fixation with what we consider adequate reduction. The patient was able to bear unprotected weight at the end of seven months (Fig. 5).

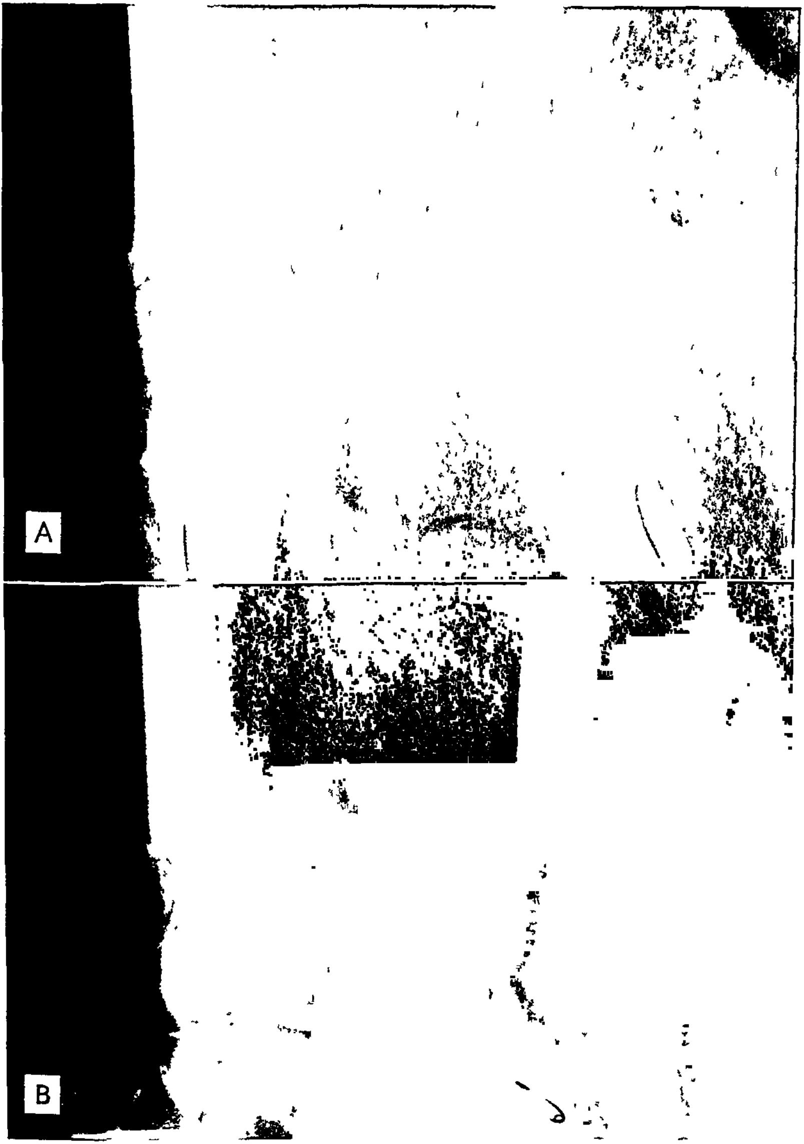
CASE 2 (E. P., aged 45).—The patient was struck by a truck on Dec. 23, 1950, and had a bilateral fracture of the femur (Fig. 6*A*). The right femur was fractured in the lower third and was treated by the use of a plate with screws; the left femur, which had a dicondylar fracture, was treated by the use of a Blade plate with adequate reduction. Operation was performed on December 28. The patient was placed in a Pearson attachment bilaterally, with Buck's extension. Sufficient callus for freedom of use was present in the supracondylar fracture at the end of four and one-half months (Fig. 6*B*). The fracture of the lower third of the femur went on to nonunion subsequently, requiring a bone graft and six additional months before weight bearing could be permitted.

CASE 3 (W. S., aged 53).—This was one of our early cases, in which the patient was treated unsuccessfully by open reduction and insertion of the screw to hold the dicondylar fractures together and the use of a Steinman pin through the tibial tuberosity for longitudinal pull and a Kirschner wire transversely through the shaft of the femur to maintain adequate reduction at the fracture site. The patient was injured in an automobile accident on Sept. 13, 1947. He was moved to Chicago, where operation was performed on October 21. The patient was treated in a spica cast for six months, without successful union. It was recommended that a bone graft be applied to the area, to which, up to the time of writing, the patient has refused to consent. He was last seen in September 1950, with a sclerosing nonunion and walking with the aid of two crutches and on some occasions a crutch and a cane (Fig. 7). This patient has only 20 degrees of knee motion. In our opinion, about 5 degrees of this motion actually comes from the site of nonunion.

CASE 4 (C. B., aged 54).—The patient, a woman, was injured in September 1948 as a result of an automobile accident in California and came under our care in December 1949 (Fig. 8*A*). Conservative management was instituted because physical therapy was desired in order to increase the muscle tone and see what range of motion could be obtained prior to operation. On Feb. 23, 1950, an iliac bone grafting operation was performed in order to fill the defect at the supracondylar area, which did not appear to have a complete union, and in June 1950 unlimited weight bearing was permitted on this extremity (Fig. 8*B*). The patient had considerable pain and discomfort in the foot because of the atrophy of the bones, the weakness of the muscles and the equinous position she had acquired during the two-year period since the accident, however, when last seen, in January 1951, she was walking quite well with the aid of a cane, was regaining confidence and muscle power in her knee and at the time of her most recent examination had 25 degrees of knee motion. There was solid bony union at the site of fracture.

CASE 5 (G. D., aged 43).—On July 11, 1950, the patient, a man, slipped off a ladder at work and sustained a supracondylar fracture of the right femur, which was treated unsuccessfully by an open reduction. Malunion resulted (Fig. 9*A*). The patient came under

Fig. 9 (G. D., Case 5).—*A*, malunion of supracondylar fracture treated elsewhere by open reduction. *B*, roentgenogram taken three and one-half months after treatment by authors' method (see text).


A
B

A
B

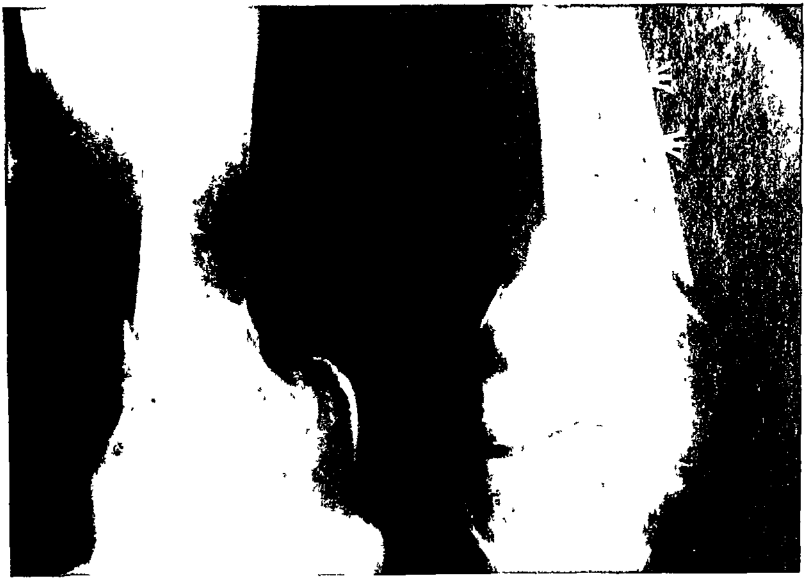


Fig. 11 (M. D., Case 7).—Supracondylar fracture of right femur in a man aged 49.

our care in December 1950, and on December 28 the protruding spur of the femur from the distal end of the proximal fragment, which protruded into the quadriceps tendon, was removed. The bone was smoothed down to allow the patella unobstructed excursion, and it was found necessary to pack a large number of bone chips into the cavity on the medial and lateral aspect of the supracondylar area, where union was incomplete. Postoperatively the man was placed in traction and received physiotherapy. Three and one-half months later he was able to walk without any form of external support (Fig. 9*B*). The ultimate range of motion was 55 degrees.

CASE 6 (R. L., aged 42).—The patient usustained a compound fracture of the supracondylar area of the femur on Dec. 4, 1949. This was debrided and treated by the use of antibiotics and Kirschner wires, one of which was placed through the tibial tuberosity for longitudinal pull and one through the proximal fragment to correct the tendency toward posterior angulation (Fig. 10*A*). After six months of conservative management it was observed that nonunion existed, and on June 6, 1950, the patient was operated on. A massive bone graft was removed from the tibia and inserted across the area of nonunion, and a Blade plate was inserted for additional support and protection. The patient was placed in skeletal traction for thirteen weeks, after which he was permitted to be up and about on a walking caliper (Fig. 10*B*). He was able to get around without any form of external support after seven additional months.

CASE 7 (M. D., aged 49).—On Dec. 20, 1949, the patient fell on a sidewalk and sustained a supracondylar fracture of the right femur. He was placed in skeletal traction, with a pin through the tibial tubercle, and was later treated in a cast. At the end of eighty days, careful examination of the frac-

Fig. 10 (R. L., Case 6).—*A*, compound supracondylar fracture of femur. *B*, taken thirteen weeks after bone graft operation.

[illegible] nonunion (Fig. 11). On May 4, [illegible] patient was operated on. A massive [illegible] graft was removed from his tibia and [illegible] across the fracture site into the [illegible] of the femur. A Blade plate was ap[illegible]. At the end of nine weeks the patient [illegible] permitted to be up and about on a brace [illegible] of crutches. The patient was [illegible] get around with unprotected weight [illegible] the end of five and one-half months [illegible] 12).

[illegible] (C. K., aged 35).—On March 29, [illegible] the patient, a woman, was involved in [illegible] accident and sustained mul[illegible], including a marked cere[illegible]. At the end of seven weeks [illegible] was referred to the office for care. [illegible] reduction was performed of the [illegible] fracture of the femur, three [illegible] used for internal fixation, one [illegible] (8.8 cm.) in length and two [illegible] (5 cm.) in length. The pa[illegible] treated postoperatively in traction [illegible] spica cast. She wore the cast [illegible] months after her discharge, obtain[illegible] appeared to be a good union. She [illegible] crutches and a walking caliper.

[illegible] 27, 1949, it was observed that the [illegible] of fracture was gradually bending and [illegible] evidence of a sclerosing nonunion was [illegible] more and more apparent (Fig. [illegible]). The patient was somewhat reluctant [illegible] further surgical treatment. On [illegible] 28, 1950, however, a massive bone [illegible] removed from the same tibia and a [illegible] inserted for additional fixation. [illegible] later the patient was permitted [illegible] about on crutches with a caliper. [illegible] solid bony union seven months [illegible] which time she was permitted to bear [illegible] any protection (Fig. 13*B*).

CONCLUSIONS

[illegible] opinion, the treatment of supra[illegible] fractures with displacement that [illegible] reduced early re[illegible] fixation for prevention [illegible] intervention with firm [illegible] early mobilization [illegible] minimal amount of [illegible] and restriction of

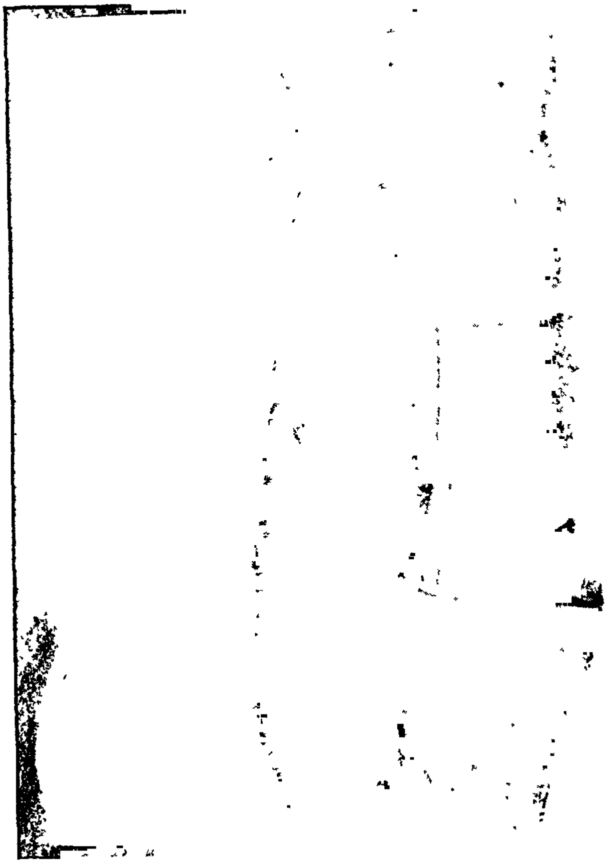


Fig. 12 (M. D., Case 7).—Condition of fracture in Fig. 11 nine weeks after bone grafting (see text).

SUMMARY

1. The authors present 2 typical cases of fresh supracondylar fracture treated successfully by the method here described.

2. With supracondylar fractures that heal by malunion or insufficient bone formation, it is important that the deformity be rounded off so as not to interfere with the patella or extension mechanism of the quadriceps tendon, and that a graft be inserted in the areas where there is insufficient bone formation to constitute a good solid union.

3. In all cases of nonunions it is important that a massive bone graft be inserted deep into the condyle of the femur for firm fixation along the shaft with additional reinforcement by a Blade plate to

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[illegible] fracture of femur due to motor accident. Patient, a [illegible] condition after open reduction, indicating sclerosing [illegible] after bone grafting procedure (see text).

A
B

insure firm fixation and maximum protection against fracturing of the graft. Knee motion can be started earlier under this regime, and the maximum amount of function is thereby preserved.

4. The method presented is not technically too difficult, and authors' experience and results have been most favorable. There has been no failure to gain union in the cases here presented.
5. In the treatment of fresh fractures, the Blade plate fixation definitely shortens the period of convalescence and acts as an excellent preventive of nonunion.

CONCLUSIONI

In casi di fratture sovracondiloidee del femore con spostamento dei frammenti che non possano essere precocemente ridotte in modo soddisfacente, una loro solida immobilizzazione cruenta è di grande importanza ai fini della prevenzione della pseudoartrosi. L'intervento precoce e la solida fissazione rendono possibile la mobilizzazione precoce del ginocchio col minimo di aderenze post-operatorie e di riduzione della mobilità.

Gli AA. presentano 2 casi tipici di fratture sovracondiloidee recenti curate con tale metodo in cui il risultato fu favorevole.

Quando poi una frattura sovracondiloidea guarisce con un callo osseo insufficiente o mal consolidato, è della massima importanza che il cerchiaggio venga eseguito in modo da non interferire con la patella o con il tendine del quadricipite, come pure che il trapianto venga innestato nella zona dove il callo osseo è insufficiente per permettere una buona e solida saldatura.

In ogni caso di pseudoartrosi è importante, ai fini di una solida immobilizzazione lungo l'asse, che un grosso trapianto osseo venga innestato profondamente nei condili femorali, aggiungendo una placca di Blade per assicurare una sicura contenzione e un massimo di protezione contro una frattura del trapianto. Con tale procedimento la mobilizzazione del ginocchio può essere iniziata molto precocemente cosicchè la funzionalità viene conservata al più alto grado.

Il metodo descritto non è tecnicamente troppo difficile e l'esperienza degli AA. è stata coronata da successo; infatti, nei casi riferiti in questo lavoro, non vi sono state consolidazioni mancate.

Nelle fratture recenti, la placca di Blade abbrevie nettamente il periodo di convalescenza ed agisce come un eccellente profilattico della pseudoartrosi.

CONCLUSIONS

Dans les cas de fractures supracondyliennes avec déplacement qui ne peut être réduit précocement, une fixation interne ferme est importante pour prévenir la non-union. L'intervention précoce et une fixation ferme rendent possible la mobilisation précoce du genou avec la quantité minima d'adhérences post-opératoires et limitation du mouvement.

Les auteurs prèsentent 2 cas typiques de fractures "froides" supra-condyliennes traités par cette méthode. Dans les 2 cas, le résultat fut un succès.

Quand une fracture supra-condylienne guérit avec mauvaise union ou insuffisance de formation osseuse, il est important que la déformation soit aplanie pour ne pas nuire à la rotule ou au mécanisme d'extension du tendon du quadriceps ou qu'une greffe soit insérée où il y a manque de tissu osseux pour permettre une union complète et solide.

Dans tous les cas de non-union il est important qu'une greffe osseuse massive soit insérée profondément dans le condyle du fémur avec ferme fixation dans la diaphyse avec réenforcement additionel par l'usage d'une plaque "Blade" pour assurer la fixation solide et protection maxima contre la fracture de la greffe. Les mouvements du genou peuvent être commencés plus tôt sous ce regime et ainsi on sauve plus de fonction.

Cette méthode n'est pas techniquement trop difficile et selon l'expérience des auteurs donne des résultats très favorables. Il n'y a pas eu de cas où l'on a raté la réunion solide.

Employee dans les fractures froides, la fixation par plaque "Blade" raccourcit positivement la periode de convalescence et agit preventivement contre les pseudoarthroses

CONCLUSIONES

En casos de fractura supracondilea con desplazamiento que no pueda ser reducida adecuadamente con oportunidad, es importante la fijacion interna firme en prevención de que union no tenga lugar La intervención y fijacion firme hace posible *la movilizacion temprana de la rodilla con* el minimo de adherencias postoperatorias y restriccion de movimientos

Se presentan dos casos tipicos de fractura supracondilea reciente tratada con este metodo En ambos casos el resultado fue satisfactorio

Cuando una fractura supracondilea cura con mala union o formación osea insuficiente, es importante que la deformidad sea redondeada, de modo que no interfiera la rotula o el mecanismo de extension del tendon del cuadrriceps, insertando un injerto en el area en donde es insuficiente la formacion ósea para permitir una buena y sólida unión

En todos los casos en que no haya unión es importante insertar un injerto óseo masivo profundamente dentro del cóndilo femoral, para fijar firmemente el tallo, con reforzamiento adicional con una placa de Blade para asegurar la fijacion firmemente, con proteccion maxima contra la fractura del injerto El movimiento de la rodilla puede iniciarse mas temprano con este regimen y preservar en consecuencia la funcion al maximo

El metodo que se presenta no es tecnicamente demasiado dificil y la experienca de los autores ha sido muy favorable No ha habido fracasos para lograr la union en los casos presentados en este articulo

Cuando se emplea para fracturas recientes, la fijacion con placa de Blade acorta definitivamente el periodo de convalecencia y obra como un excelente preventivo de la falta de union

CONCLUSOES

Em casos de fratura supracondilar com deslocamento que não pode ser reduzido precoce e adequadamente, fixação interna firme e importante na prevenção de *ausencia de união Intervenção precoce e* fixação firme tornam possivel a mobilização precoce do joelho com minima quantidade de adesões pos operatorias e restrição de movimentos

Os autores apresentam 2 casos tipicos de fratura supracondilar recente tratados por este metodo Em ambos os casos o resultado foi satisfatorio

Quando uma fratura supra condilar resulta em mal união ou insuficiente formação ossea e importante que a deformidade seja arredondada de modo a não interferir com a rotula ou com o mecanismo de extensão do tendão do quadriceps e que um enxêrto seja inserido nas areas onde ha formação de osso insuficiente para permitir uma bôa e solida união

Em todos os casos de não união e importante que um enxêrto osseo macisso seja aplicado profundamente dentro do condilo do femur para firme fixação ao longo do eixo, com adicional refôrco de lamina de aço para assegurar firme fixação com proteção maxima contra fratura do enxêrto A movimentação do joelho pode ser iniciada precocemente sob essas condições e em consequencia preservar a maxima quantidade de função

O metodo apresentado não e tecnicamente muito dificil e a experiencia dos autôres tem sido a mais favoravel Não tem havido fracasso para conseguir união nos casos apresentados neste artigo Quando usada para fraturas recentes a fixação por lamina de aço definitivamente encurta o periodo de convalescença e age como excelente preventivo de não união

ZUSAMMENFASSUNG

In Faellen von suprakondylaeren Oberschenkelbruechen, in denen die Fragmentverschiebung nicht fruehzeitig ausgeglichen werden kann, ist eine feste innere

Fixierung zur Verhuetung von Heilungsversagern wichtig. Ein fruehzeitiges Eingreifen und feste Fixierung ermoeglichen fruehe Bewegung des Knies mit einem Mindestmass von postoperativen Verwachsungen und Bewegungsbehinderungen.

Die Verfasser stellen zwei typische mit dieser Methode behandelte Faelle von frischen suprakondylaeren Bruechen vor. In beiden Faellen war das Ergebnis erfolgreich.

Wenn ein suprakondylaerer Bruch mit schlechter Vereinigung der Fragmente oder unzureichender Knochenbildung heilt, ist es wichtig, die Deformierung abzurunden, damit keine Stoerung der Kniescheibe oder des Streckmechanismus der Quadrizepssehne eintritt; es ist ferner wichtig, dass ein Knochenspan in die Stellen, wo unzureichende Knochenbildung vorliegt, eingepflanzt wird, um eine gute solide Vereinigung zu ermoeglichen.

In allen Faellen von fehlender Knochenvereinigung ist es wichtig, einen massiven Knochenspan tief in den Oberschenkelhoecker einzupflanzen, um eine feste Fixierung in der Richtung des Schaftes zu erreichen; darueber hinaus soll eine Verstaerkung mit Hilfe einer gewinkelten Metallplatte angelegt werden, um neben der soliden Fixierung die bestmoegliche Vorbeugung eines Bruches des Knochenspans zu erzielen. Mit diesem Verfahren kann die Bewegung des Knies fruehzeitiger begonnen und somit die Erhaltung einer maximalen Funktion gesichert werden. Die angegebene Methode ist technisch nicht zu schwierig und die Verfasser haben mit ihr aeusserst guenstige Erfahrungen gemacht. In den in der vorliegenden Arbeit dargestellten Faellen sind Heilungsversager nicht vorgekommen.

In Faellen von frischen Bruechen verkuerzt die Fixierung mit Hilfe der gewinkelten Metallplatte sicherlich die Zeit der Rekonvaleszenz; gleichzeitig dient sie als ausgezeichnetes Vorbeugungmittel einer nichtknoechernen Vereinigung.

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To Our Contributors Abroad

We regret to announce that under current publishing conditions it has become impossible for the *Journal* to arrange the translation into English of original articles submitted in other languages. With the single exception of articles written specifically for the *Seccion en Espanol*, which, of course, do not require translation, we must therefore request that all articles henceforward be submitted in the English language. Prompt publication can be greatly facilitated if all manuscripts are typed in double or triple space to allow room for editorial corrections. Your cooperation will be deeply appreciated.

Bursitis and Myofascitis

Treatment by Local Injections of Cortisone: a Preliminary Report

EGMONT J ORBACH, M D, F I C S
NEW BRITAIN CONNECTICUT

THE clinical response to local injections of cortone®* was investigated in various types of rheumatic involvement, ulcerative lesions and dermatologic conditions.

The study was prompted by the report of Koff and his associates[1] on injections of cortisone in cases of iritis. Seventy-nine cases were investigated in the period between December 1950 and September 1951.

It became evident during this study that bursitis and myofascitis could be managed successfully by local cortone infiltrations. These two groups are described in this paper.

From the physiopathologic standpoint, cortisone suppresses inflammatory reactions. Taubenhaus and Amromin[2] and also Conn[3] observed that cortisone has an inhibiting effect on fibroblasts and collagen formation. Carlisle and his coworkers[4] have photographically recorded the depressant action of the compound on connective, lymphatic, granulation and bone tissue. Inhibition of the Koch phenomenon and of the tuberculin reaction by this drug, as demonstrated by these investigators, is notable.

In the present series, local injections of cortone were given four to fourteen days apart to a total of 55 patients with the following conditions: bursitis, calcifying tendinitis and myofascitis.

It was felt that to obtain optimum local effects the affected tissues should be infiltrated with a large volume of material, and for this reason the standard suspension of cortone was diluted in all but a few cases. Because of the possibility of pain on infiltration, a weak solution of procaine hydrochloride was used as the diluent. The strength of the solution was not permitted to exceed 0.25 per cent (in order to keep therapeutic effects from the procaine to the minimum). Diluting the suspension did not impede the action of the cortone. Instead, by permitting decreased doses, it prevented occurrence of such side effects as sodium retention, hypertension, hyperglycemia, hypopotassemia, and the Cushing syndrome, which may follow systemic administration.

The following technic was used: 12.5 to 100 mg. of cortone suspension was diluted with 1 to 20 cc. of a 0.125 to 0.25 per cent solution of procaine hydrochloride. The material was injected locally into the region of greatest pain by means of a 22-gauge needle 2 to 4 inches (5 to 10 cm.) long. The area involved was infiltrated thoroughly by partial withdrawal of the needle and redirection of its course as often as necessary during an injection.

Representative cases from each group are reported in the body of this paper. The tables contain pertinent data noted in all cases studied.

In the treatment of the pain syndrome the evaluation of results is often difficult, because objective evidence on which to base a conclusion frequently is absent. In cases of acute bursitis with limitation of motion, an increase in active and passive mobility of the shoulder after treatment can be considered an indication of improvement. However, in cases of chronic bursitis or myofascitis without restriction of motion, pain is the only indicative symptom and the subjective response of the patient the only criterion of improvement. In the following section, patients showing sustained early or dramatic re-

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*Cortisone is manufactured under the trade-mark name Cortone by Merck & Co, Inc, Rahway, N J.

Submitted for publication Nov. 8, 1951.

mission of symptoms have been classified as "improved", while "fair" denotes the results in those whose improvement either was partial or failed to exceed that obtained previously with other therapeutic measures. The results in patients not responding to cortisone therapy have been classified as "poor".

1. *Bursitis and Calcifying Tendinitis.*—Twenty-one patients, 13 women and 8 men ranging in age from 30 to 70 years, were treated in this group. In 1 patient the retrocalcaneal bursa was involved; in 14, the subacromial; in 1, the popliteal; in 3, the trochanteric; in 1, the radial, and in 1, the ischiogluteal. The number of injections varied from one in 9 cases, two in 5, three in 1, and four in 4, to six in 2. Initially, cortone was infiltrated in small amounts ranging from 12.5 to 25 mg., but as the study progressed it was found that relief could be obtained more promptly with doses of 25 to 100 mg. Good results were observed in 12 patients and partial improvement in 6. Three did not respond to therapy.

The time required to obtain improvement varied from one day to three months. Patients with subacromial bursitis were instructed to keep the arm in abduction and to practice shoulder exercises.

CASE 1.—Mrs. F. B., aged 49, for four years had had intermittent attacks of excruciating pain in the right hip, radiating toward the knee. One year prior to admission, in a severe attack lasting two weeks, physiotherapy and four procaine infiltrations had been required to relieve the pain. Roentgen examination at the onset of the reported recurrence revealed calcification above the trochanter. Temporary relief from pain was produced by roentgen therapy, during which a total of 806r was directed over the calcification. Two weeks later the patient was confined to bed because of the severity of the pain in the right hip. Cortone therapy was instituted, 50 mg. in 20 cc. of 0.125 per cent procaine hydrochloride being infiltrated into and around the trochanteric [illegible]. The next morning the patient was free from pain, but there was some soreness at the site of injection. The procedure was repeated on the fourth and fifth day with 50, 75 and finally 100 mg. of cortone. Postinjection soreness resulted from the last injection but was followed by complete relief of pain.

CASE 2.—Mr. A. C., aged 30, had severe intermittent pain in the left hip for three years. When examined after two days of severe pain, the lower left extremity was kept in adduction, with the body drawn toward the affected hip. The area over the left trochanter was swollen and extremely tender, and the cutaneous temperature in this region was increased. Walking was extremely painful, and the patient limped. Cortone, 12.5 mg. in 20 cc. of 0.125 per cent procaine hydrochloride was cautiously infiltrated into the painful area. A needle 4 inches (10 cm.) long was used to permit an inferior approach. Mild postinjection soreness appeared a few hours later, but dramatic relief occurred the next day. The patient could walk without limping and with full motion of the left hip. Although it was not necessary, a second infiltration was performed that day at his request.

CASE 3.—Mrs. E. F., aged 59, had pain in the right shoulder for four months, increasing in severity until it became acute four days before treatment. The area below the right acromion was swollen and extremely painful on palpation; the cutaneous temperature was 1 degree (C.) higher than on the left side. Shoulder muscle spasticity limited active elevation of the arm to 150 degrees. The right subacrominal bursa was infiltrated with 25 mg. of cortone in 10 cc. of 0.125 per cent procaine hydrochloride, but no relief was obtained. Three days later the procedure was repeated with 50 mg. of cortone in 10 cc. of 0.25 per cent procaine hydrochloride, but again there was no improvement. During the following week roentgen examination revealed a small calcification at the greater tubercle, and 100 mg. of cortone in 10 cc. of 0.25 per cent procaine hydrochloride was infiltrated into and around the subacromial bursa. Satisfactory relief was obtained four days later. When the final infiltration with 100 mg. of cortone was given, full range of motion was present in the shoulder.

The shoulder again became painful six weeks later, but motion was not restricted. As the patient preferred not to have local injections repeated at that time, roentgen therapy was given, with good results. Shoulder pain recurred in two months and cortone infiltrations were reinstituted. In this case cortone therapy was effective in the acute phase

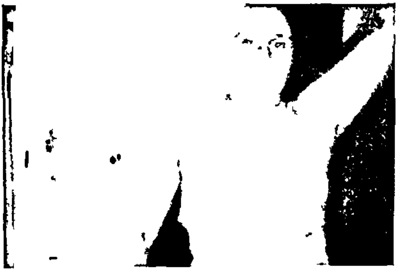

Fig 1 —Right subdeltoid bursitis with frozen shoulder Condition before injection of 50 mg of cortisone in 10 cc of 0 25 per cent procaine hydrochloride

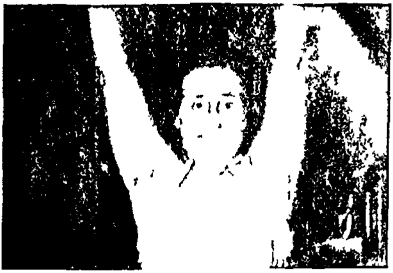

Fig 2 —Patient clinically cured of right subdeltoid bursitis after local injection of 50 mg of cortisone into painful area

of the bursitis but not in the chronic stage

CASE 4 —Mr W G, aged 60, had severe pain in the left arm and hand The temperature was 100 F The patient appeared ill The left hand was edematous, and the patient could not dress himself The area of the greater tuberosity of the left shoulder was swollen, and roentgen examination revealed a soft shadow in this area Cortone, 100 mg in 10 cc of 0 125 per cent procaine hydrochloride, was infiltrated into and around the subacromial bursa The patient was instructed to keep the hand elevated and to do shoulder exercises For twenty-four hours after the injection he was unable to move the shoulder joint because of increased soreness This was followed first by diminished pain and then a "cracking" sensation in the shoulder, after which he could move the joint without difficulty On the next day there was stiffness but no pain in the shoulder, and the patient was able to dress himself without assistance Active and passive motion of the shoulder was normal The edema of the hand had completely disappeared

CASE 5 —The case of Mrs S K, aged 40, is reported because cortone proved ineffective and therapeutic results were obtained by systemic administration of antibiotics This patient complained of severe pain in the right heel of eight weeks' duration The soft tissue on either side of the achilles tendon was swollen and tender, and the deltoid ligament was painful on palpation Urethrocervical smears gave negative results for gonococci Roentgen examination revealed no pathologic change Cortone, 12 5 mg in 10 cc of 0 25 per cent procaine hydrochloride, was injected into the area anterior to the achilles tendon on two occasions, without relief Antibiotic therapy was effective at a later stage

CASE 6 —Mrs S P, aged 58, had severe immobilizing pain in the right shoulder for two weeks Cortone, 50 mg in 10 cc of 0 25 per cent procaine hydrochloride, was injected into the area of greatest pain Four days later the patient was completely pain-free and could move the shoulder joint freely Only slight postinjection soreness was present

CASE 7 —Mr S K, aged 59, had acute subacromial bursitis of one week's duration The arm was completely immobile in the shoulder joint Cortone, 50 mg in 10 cc of 0 25 per cent procaine hydrochloride, was injected into the acutely inflamed area The patient was instructed to elevate the arm gradually and to practice shoulder exercises Forty eight hours later he presented himself fully recovered, without pain and with full range of motion There was no postinjection soreness

CASE 8 —Mrs B T, aged 47, showed the same clinical picture as was observed in Case 7 She received the same dose of cortone and was fully relieved after seventy-two hours No postinjection soreness was reported

CASE 9 —Mrs S C, aged 57, complained of severe pain in her leg, which could not be moved actively The condition had persisted for two weeks The area of the right trochanter appeared swollen and tender, and the cutaneous temperature was elevated Cortone, 50 mg in 10 cc of 0 25 per cent procaine hydrochloride, was injected into the painful and inflamed area On account of the severe pain, 100 mg of demerol was given The patient reported complete relief after five days

2 *Myofascitis* —This group consists of 34 cases The patients were 24 women and 10 men ranging in age from 21 to

TABLE 1.—*Bursitis Treated with Local Cortone Injection*

Case	Age	Type of Bursitis and Degree of Involvement	Dosage: Cortone Mg.	Dosage: Procaine Solution, 0.125 to 0.25%, Cc.	Region Infiltrated	No. of Injections	Duration of Condition: Before Therapy	Duration of Condition: After Therapy	Results
1 Mrs. F. B.	50	Trochanteric: acute	50 75 100	20 20 8	About trochanter major muscle	2 1 1	2 months	4 weeks	Improved
2 Mr. A. C.	30	Trochanteric: acute	12.5 25	20 20	About trochanter major muscle	1 1	2 days	1 day	Improved
3 Mrs. E. F.	59	Subacromial: acute flareup	25 50 100	10 10 10	Subacromial	1 1 2	4 months	2 weeks	Fair
4 Mr. W. G.	60	Subacromial: b. temp. 100 F.: immobile shoulder: edema of hand: acute	100 50	10 10	Subacromial	2 1	1 week	2 days	Improved
5 Mrs. S. K.	40	Retrocalcaneal: chronic	12.5 25	10 10	Retrocalcaneal: injections	1 1	8 weeks		Poor: relieved by antibiotic therapy
6 Mrs. S. P.	58	Subdeltoid: immobile shoulder: acute	50	10	Subdeltoid	1	2 weeks	4 days	Improved
7 Mr. S. K.	39	Subacromial: immobile shoulder: acute	50	10	Subacromial	1	1 week	2 days	Improved
8 Mrs. B. T.	47	Subacromial: immobile shoulder: acute	50	10	Subacromial	1	1½ weeks	3 days	Improved
9 Mrs. S. C.	57	Trochanteric: acute	50	10	Trochanteric area	1	2 weeks	5 days	Improved
10 Mrs. M. G.	41	Subacromial: acute	12.5	5	Subacromial	1	2 days	2 days	Improved
11 Mr. J. I.	49	Popliteal: chronic	25	10	Popliteal space	1	2 weeks	2 weeks	Fair
12 Mrs. F. G.	73	Subacromial: chronic	25 25 50	5 10 10	Subacromial	2 1 1	2 years	3 months	Fair
13 Mrs. F. F.	58	Radial: chronic	25 50	5 10	Around radial head	1 1	2 months		Poor
14 Mrs. H. D.	71	Subacromial: chronic	50	10	Subacromial	1	2 weeks	1 week	Improved

TABLE 1. (*Cont'd*)—*Bursitis Treated with Local Cortone Injection*

Case	Age	Type of Bursitis and Degree of Involvement	Dosage		Region Infiltrated	No. of Injections	Duration of Condition		Results
			Cortone, Mg.	Procaine Solution, 0.125 to 0.25%, Cc.			Before Therapy	After Therapy	
15 (Mr. S. M.)	60	Subacromial	25 50	10 10	Greater tuberosity of humerus	4 2	4 months	2 months	Fair
16 (Mrs. A. T.)	41	Subacromial calcification, chronic	50	10	Subacromial	2	2 years off and on	3 weeks	Fair
17 (Mrs. P. R.)	45	Subacromial, calcification	25 75 100	10 10 10	Subacromial	2 1 1	3 years	3 weeks	Fair
18 (Mrs. C. M.)	46	Subacromial, chronic	20 25 50	5 10 10	Painful area greater tuberosity	1 3 2	3 years		Poor
19 (Mrs. H. S.)	72	Ischiogluteal; subacute, diabetes	25	10	Painful area os ischii	1	6 weeks	2 weeks	Improved
20 (Mrs. H. A.)	55	Subacromial, acute	25	1	Greater tuberosity	2	1 week	2 days	Improved
21 (Mr. Z. B.)	56	Subacromial; immobile shoulder; acute, edema of hand	50	10	Greater tuberosity	1	4 days	1 day	Improved

82 years, with myofascitis involving such muscles as the metatarsal, gluteal, sacrospinal, latissimus dorsi, trapezius, rhomboid and subscapular, the lumbodorsal fascia and the iliotibial tract. The number of injections varied from one in 12 patients, two in 10, three in 3, four in 5, five in 2 and six in 1, to ten in 1. Pain was present for one to sixteen weeks before treatment. Seventeen patients recoverd completely in from one to fifty-six days, and fair results were obtained in 9 cases. Therapeutic response was poor in 8 patients; in 2 of these osteoporosis and a herniated disc were discovered to be the underlying causes of the symptoms.

In 3 patients small hard nodules could be felt near the sacroiliac joints, and palpation elicited considerable pain. As the tentative diagnosis of herniated fat could not be verified without resort to operation or biopsy, these conditions were classified as fibrositis. Within four to fourteen hours after injection several patients experienced accentuation of pain, which was followed in turn by dramatic relief.

Acute fascitis and myositis responded more readily to therapy than did the chronic forms of these conditions.

CASE 1.—Mrs. F. C., aged 48, had pain in the gluteal region of two months' duration. Examination revealed pain radiating along the course of the sciatic nerve, with exquisite tenderness over the gluteal muscle, and of the fascia in the region of the sacroiliac joint; indurated nodules could be palpated through a thick pad of fat. A diagnosis of acute lumbodorsal fascitis was made. One injection of cortone, 25 mg. in 20 cc. of 0.25 per cent

SOME CLINICOPATHOLOGICAL OBSERVATIONS ON SCLEROMA*

By PROFESSOR HANDOUSA and ANWAR M. ELWI (Cairo)

SCLEROMA is a disease characterized by chronic specific inflammatory lesions of the upper respiratory tract. Recent investigations of Kauffmann (1949), and Levine (1951) appear to confirm the ætiological relation between *klebsiella rhinoscleromatis* and scleroma. The disease has a wide distribution in Central and Southern Europe but it is established now that no part of the world is immune. We see in the Kasr El-Aini Hospital an average of 40 cases per year.

In the following account we shall forward some observations on certain histopathological and clinical aspects of the disease which to our knowledge did not receive enough attention or have not been reported beforehand.

Histopathology of Scleroma

The microscopical picture of scleroma is that of a granulomatous inflammation which develops in the subepithelial tissues of the upper respiratory passages.

At an early stage the granuloma appears rich in young vascular spaces separated from each other by a cellular infiltrate which is essentially made of Mickulicz cells, large mononuclear cells, plasma cells, and lymphocytes. The relative number of these cells is variable but commonly, the Mickulicz cells are the chief component of the infiltrate. Mickulicz cells are large in size, sharply outlined and have abundant vacuolated or foamy cytoplasm. The nucleus is small, centrally placed, and often pyknotic. Within the vacuoles of Mickulicz cells, Gram-negative diplobacilli of *K. rhinoscleromatis* may be demonstrated. Transitional stages may be seen between large mononuclear cells and fully developed Mickulicz cells. Occasionally the plasma cells degenerate and change into oval acidophilic homogeneous anuclear masses known as Russell bodies. As the lesions get older, the vascularity of the granuloma decreases gradually while the cellular infiltrate gives place to proliferating fibroblasts but the resulting fibrous mass remains fairly infiltrated with lymphocytes and plasma cells.

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In our experience, the microscopic picture of the lesions is fairly specific and diagnostic of the disease; the one pitfall which one may encounter is the occasional case of nasal leprosy. Lepra cells may simulate the Mickulicz cells to such a degree that the picture of the leprotic granuloma as seen in routine sections stained with hæmatoxylin and eosin becomes indistinguishable from that of scleroma. This is not astonishing because both of the lepra cells and Mickulicz cells are derived from large mononuclear cells. Stains for Gram-negative *K. rhinoscleromatis* and acid-fast *Mycobacterium leprae* in sections are not successful and helpful in all cases This problem directed our attention to the study of the foamy nature of the Mickulicz cells of scleroma.

A close study of the literature did not reveal to us any explanation of the mechanism of the change which takes place in the cytoplasm of large mononuclear cells as they are transformed into Mickulicz cells.

For this, we planned to carry out a preliminary histochemical study of the lesions of scleroma.

Methods

Histological sections were prepared from biopsy material and stained with (1) routine hæmatoxylin and eosin, (2) periodic acid-Schiff (Hotchkiss, 1948), (3) acid hæmatein (Baker, 1946), (4) colloidal aqueous Sudan III (Govan, 1944), and (5) Millon's reaction as modified by Bensley and Gersh (1933).

Results and Discussion

Our plan was to investigate the possibility that the vacuolated appearance of the Mickulicz cell was due to the loss of some abnormal cytoplasmic substance during the histological processing of the tissue. As regards the origin of such an abnormal substance, two possibilities presented themselves. One possibility is the mucoid substance which makes the capsule of *K. rhinoscleromatis* (Topley and Wilson, 1956); the other one is a carbohydrate, fat, or protein substance which is freed from the usual complex physical bonds with other cytoplasmic components as a result of some cellular metabolic disturbance caused by the organisms.

The histochemical stains which we chose to apply on material of scleroma are known to demonstrate a wide range of carbohydrate, fat, and protein substances (Pearse, 1954). The periodic acid-Schiff stain (PAS) as described by Pearse, gives a positive result in the presence of polysaccharides, mucopolysaccharides, muco- and glycoproteins, glycolipids, unsaturated lipids and phosphatides. Govan's Sudan III method stains neutral fats and lipins while Baker's acid hæmatein is specific for ester phosphatides. Millon's reaction is positive for the protein molecules of the hydroxylphenol group such as tyrosine.

The Mickulicz cell gave a negative result in all these tests (Figs. 1, 2, 3). Therefore we concluded that neither the capsular mucoid substance of the organisms accumulates in sufficient amounts that can be demonstrated by the PAS technique, nor that the cellular metabolism of carbohydrates, fats, and proteins is interfered with to the extent that free stainable substances such as enumerated before show themselves.

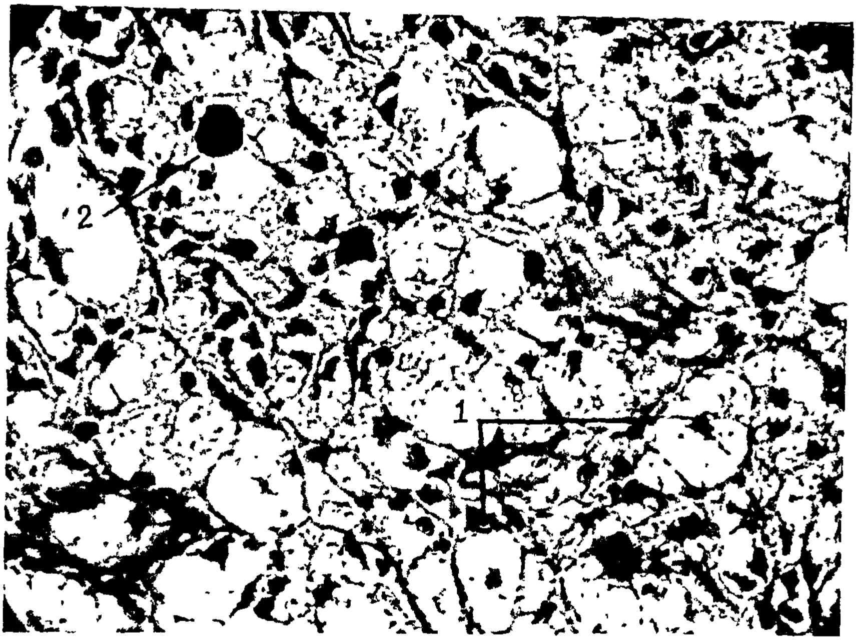

FIG. 1.
Photomicrograph of section stained with PAS. Note: (1) Negative vacuolated Mickulicz cell. (2) Russell body strongly positive. × 540.

These conclusions suggested to us that the change in the Mickulicz cells may be the result of a disturbance of the water exchange between the cells and tissue spaces. Opie (1949), and others forwarded the evidence that the intracellular fluids are hypertonic. Accepting this evidence, Robinson (1952) suggested that the water balance of the cell is governed by its respiratory activity, some of the energy released by the latter process is used for active transportation of water out of the cell and the prevention of its intracellular accumulation. He showed that poisoning of the respiratory apparatus of the cell was followed by a decrease of the oxygen consumption and a proportionate decrease of the energy released by the oxidation process. As a result of this, water accumulated inside the cells which became distended. On this basis, we offer the opinion that the phagocytosed *K. rhinoscleromatis* injure the respiratory apparatus of the large mononuclear cells in some way or other and cause the intracellular

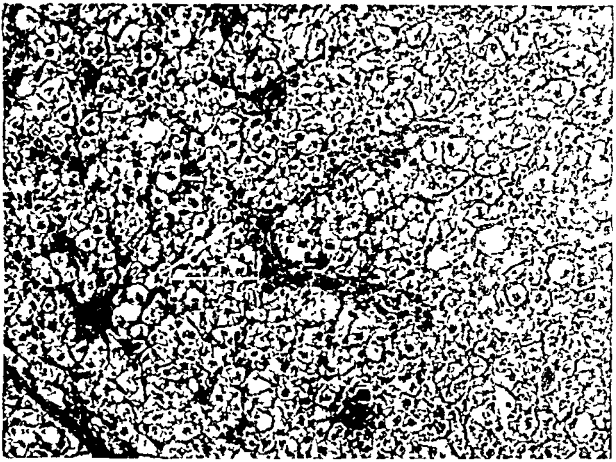

FIG 2

Photomicrograph of section stained with acid hæmatin Note (1) Negative Mickulicz cells and (2) Positive red cells in vascular trabeculae The Mickulicz cells showed in section greenish yellow granularity ×240

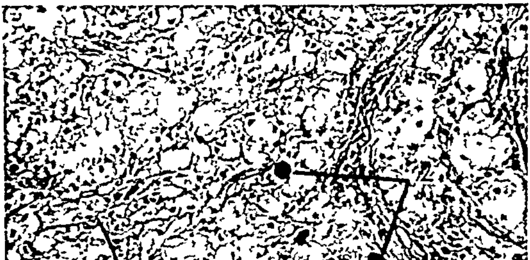

FIG 3

Photomicrograph of section stained for Millon's reaction Note (1) Negative Mickulicz cell in contrast to (2) Strongly positive Russell body ×270 accumulation of water in the way described by Robinson. The distension and the vacuolated or foamy appearance of the cytoplasm of Mickulicz cells would be due to the increase of water in the intermicellary spaces of the cell proteins, that is, the change is a sort of hydropic degeneration induced by cell injury caused by phagocytosed bacilli. With such a state of affairs one would not expect to find a histochemical staining that would characterize Mickulicz cells.

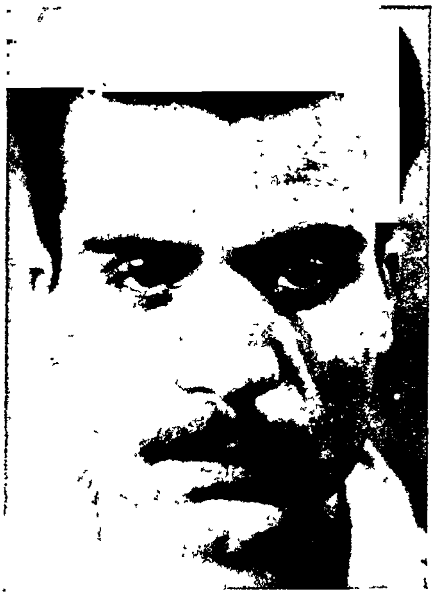

FIG. 4.

Note the tumour-like mass of the left lateral wall of nose which was the only demonstrable manifestation of rhinoscleroma in the case.

Clinical Features of Scleroma

The main clinical features of scleroma have been described both in the current literature and standard text-books, and are well known especially to those who are familiar with the disease.

At the stage when the granuloma is fully developed, the infiltrate is known to take a diffuse or a nodular form which is usually bilaterally represented. The nodular form is presented by multiple, well-localized small masses, and covered by intact mucosa in both sides of the nose. However, we came across cases of scleroma which were manifested by a single large tumour-like mass alone; or in association with other scleroma manifestations.

These tumour-like masses may (1) arise in connection with the nasal frame and either show mainly on the external surface or the inside in the

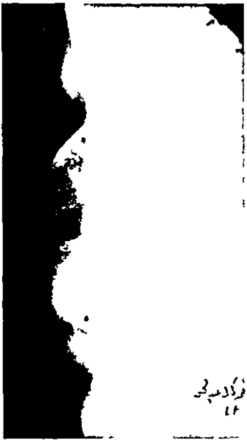

Fig 5

Radiogram of left lateral nasal wall Note pressure atrophy of the left nasal bone with surrounding secondary reaction

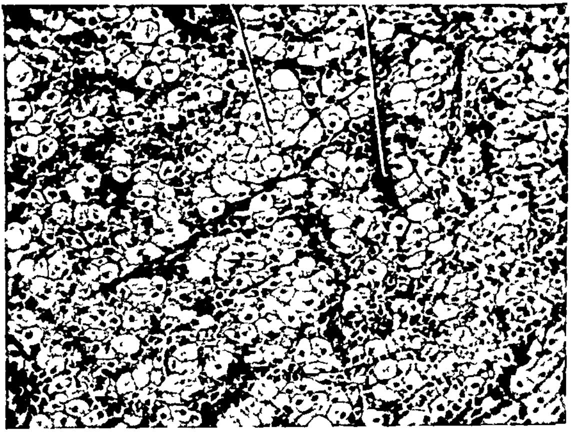

Fig 6

Photomicrograph of mass in Case No 1 Note (1) Mickulicz cell, (2) vascular spaces nasal airway; they may arise also from (2) the septum, (3) nasopharynx, or (4) larynx.

Nasal cases. We have come across 5 cases of scleroma of the nose which were manifested by a single "tumour-like" mass alone, and 8 cases in which the mass was associated with other manifestations of scleroma.

The tumour-like mass in its site, appearance and feel induces the rhinologist to think of the diagnosis of a tumour especially in the absence of other manifestations of scleroma. Some of these cases have been given the provisional diagnosis of chondroma, malignant tumour, or gumma until the routine investigations revealed that the mass was a typical scleroma. The following cases illustrate our point.

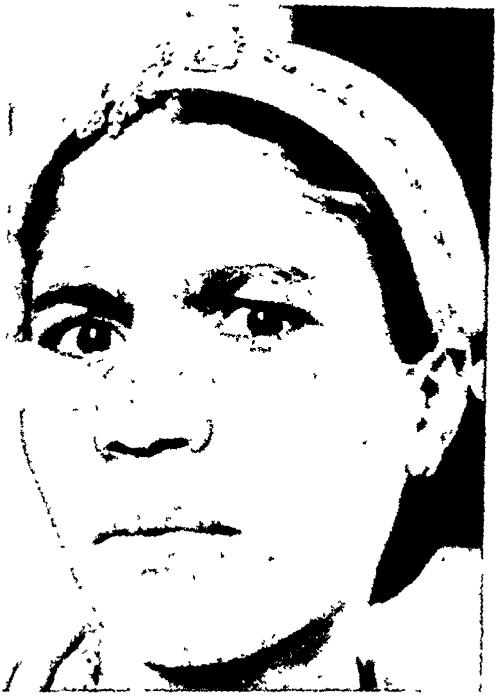

FIG. 7.

Case No. 2. Note a tumour-like mass in the right lateral nasal wall In this case other scleroma manifestations were demonstrable in the nose and larynx.

CASE I. A.A., a male, aged 40 years. The only complaint was the levelopment of a slowly and progressively enlarging mass in the left lateral wall of the nose (Fig. 4). The mass was subcutaneous but fixed to the underlying bone; it felt like a mass of cartilage. A corresponding bulge was seen in the left nasal passage but the nasal mucosa was intact and there was no demonstrable sign in the nose, nasopharynx or larynx which might suggest its scleromatous nature. Radiological examination demonstrated pressure atrophy of the underlying bone with thickening around (Fig. 5). At operation, the mass was not encapsulated and was found to be breaking down in its deep part. Microscopical examination of the mass revealed the picture of scleroma (Fig. 6).

In this case scleroma is manifested only by a single tumour-like mass, a feature which may mislead the rhinologist especially in countries in which the disease is not endemic. It also proves the subepithelial development of the disease.

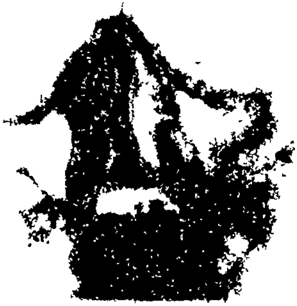

FIG 8

Radiogram of mass in Case No 2 Note the clear radiological definition and localization of the mass in the fibrotic stage

CASE 2. F.R.O., a female aged 30 years. She complained of a slowly growing mass of the right outer wall of the nose of about one year duration (Fig. 7). The mass was again subcutaneous and fixed to the deep structures. It bulged in the corresponding nasal passage and felt like a fibroma. However, the nasal mucosa was covered by scabs, the pharynx was dry and granular, and the patient had a hoarse voice. Examination of the larynx revealed a fairly large granuloma in the right ventricle. Biopsy examination proved that both of the nasal and laryngeal masses were scleromatous. Radiological examination of the nasal mass is interesting as it shows localization and extensive fibrosis (Fig. 8). This is an example of scleroma which presented itself in the form of a single tumour-like mass in relation to the nasal frame but was associated with other scleroma manifestations, in the nasal passages, pharynx and larynx.

In three cases, the patients complained of slowly progressive one-sided nasal obstruction. On examination a single tumour-like mass was seen in the obstructed side arising from the lateral nasal wall in the region of the inferior or middle turbinate bone, and looked like an angioma or malignant disease. In none of these cases were any of the other clinical manifestations of scleroma seen, though biopsy examination showed the picture of scleroma in all of them.

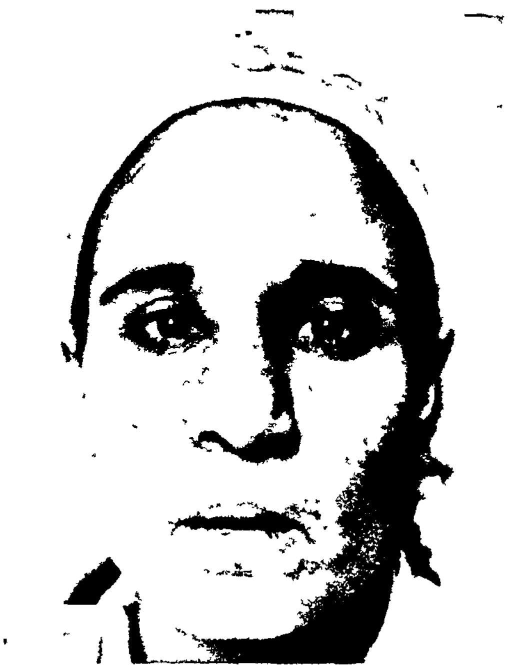

Fig 9

Photograph of a scleroma mass in relation to the root of the nose which was very suspicious of being syphilitic

Three other similar cases were seen on the nasal septum, each presented a bleeding tumour-like mass arising from the septum. The earliest case which the senior author had seen looked like an angioma, but biopsy examination by the late Professor A. F. Bernard Shaw proved it to be scleroma.

We had 2 cases in each of which there was a tumour-like mass arising in connection with the nasal frame; the mass was strongly suspicious of being syphilitic in nature, because the serological reactions were positive and the radiological examination presented the picture of periostitis. However, biopsy proved the mass to be scleroma in nature (Figs. 9 and 10).

Nasopharyngeal cases. The nasopharyngeal cases were of particular interest because they simulated closely in their position, symptoms, appearance, and feel, nasopharyngeal fibroma, the treatment of which is much more serious and needs special precautions, while a scleroma mass requires a much simpler treatment. We met with three cases of this type, one of which was arising from the roof of the nasopharynx and the two others from the nasopharyngeal surface of the palate. They were all so large as to fill the nasopharynx. The covering mucosa was intact, greyish-white in colour, and shining with dilated vessels running on the surface; the consistency was firm. In each case, the tumour-like mass was associated with other scleroma manifestations. In one case, the nasal mucosa was atrophic; in the second it was ulcerated and covered by scabs, and in the last it was granular. Biopsy examination of the tumour-like mass of the three cases disclosed the picture of scleroma.

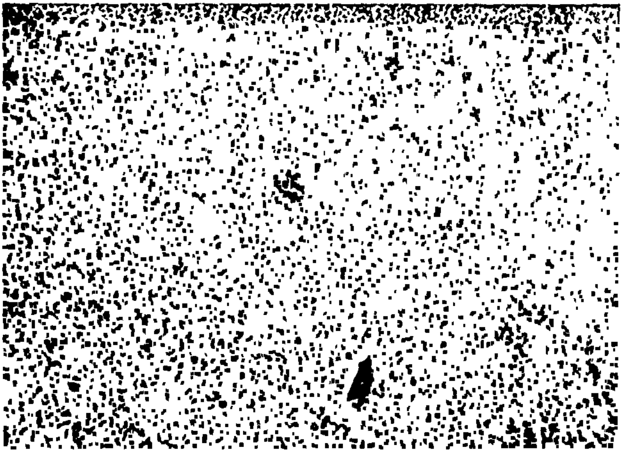

FIG 10

Photomicrograph of scleroma mass seen in Fig 9 The microscopic picture appears somewhat different from the usual one because Mickulicz cells (1) appear scanty, while plasma cells and lymphocytes make the biggest portion of the infiltrate

Laryngeal cases. In the larynx, the disease is almost always secondary to nasal scleroma. During 25 years of practice, the senior author has met with only three cases of laryngoscleroma in which there was no demonstrable signs of scleroma in the nose, nasopharynx or pharynx.

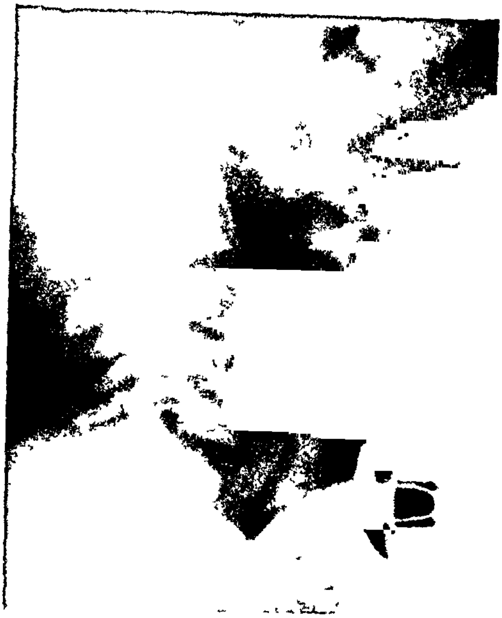

FIG. 11

Radiogram of a case of laryngoscleroma Note: (1) the almost complete occlusion of the larynx, and (2) extensive narrowing of the trachea which was uniform all through.

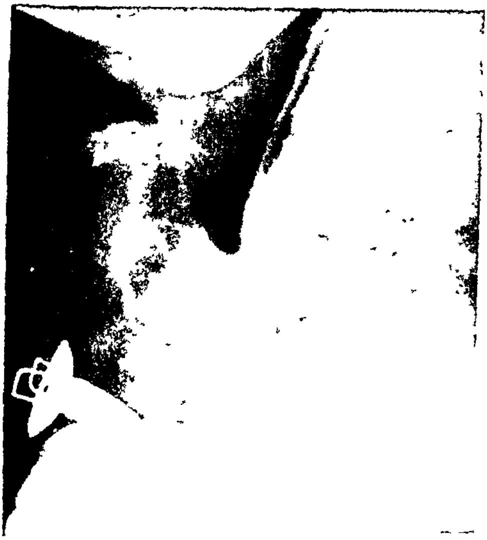

FIG. 12.

Radiogram of a case of laryngoscleroma. Note: (1) the narrowing of the tracheal lumen is uniform all round in one section of the tube but it varied at different levels. (2) The picture in this radiogram gave the false impression of increased thickness of the tracheo-vertebral space. This has been excluded by a barium swallow.

The site of choice for the lesion is the "subglottic region". Crusts may form over the glottis. In the late stages of the disease, a fibrous stricture is formed below the level of the vocal cords and as a result the airway is markedly reduced. The vocal cords often remain freely mobile.

Unilateral affection of the larynx had been seen in the form of a granuloma of the ventricle (in 5 cases), and thickening and fixation of one vocal cord with or without a granuloma in relation (in 6 cases). The latter form closely simulated malignant disease in its clinical appearances.

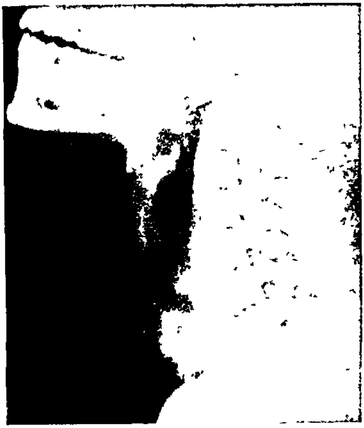

FIG 13.

Radiogram of laryngo-tracheo-scleroma. Note the shadow of well localized masses in the trachea

Tracheo-scleroma. The tracheal lesions in our group of cases have been always associated with laryngoscleroma. It has been claimed that the lesions in the trachea could be primary but we have never seen one. The lesion leads to progressive stenosis of the trachea. Direct and indirect view of the tracheal lumen is usually very difficult to attain owing to the associated laryngeal stenosis in advanced cases. For this reason the presence of a lesion in the trachea, its localization and the confirmation of its nature by biopsy has been difficult or impossible to demonstrate by endoscopy.

However, scleroma lesions in these advanced cases have been found demonstrable radiologically especially in the cervical portion of the trachea. This encouraged us to study their radiological features as well as the previous data in 14 consecutive cases with the following results:

1. The lesion was always extensive in the *mucosa* involving a long length of the trachea or all of it, causing thickening with secondary narrowing of the air column. We were never sure of the spread of the disease to the cartilage rings, and there was no spread at all outside the tube. The X-ray *shadow* of the thickened mucosa was sometimes seen diffuse with the shadow of the structures lying behind the trachea giving the false impression of "widening of the tracheo-vertebral space", but this was ruled out easily by examining radiograms taken while the patient was swallowing an opaque meal.

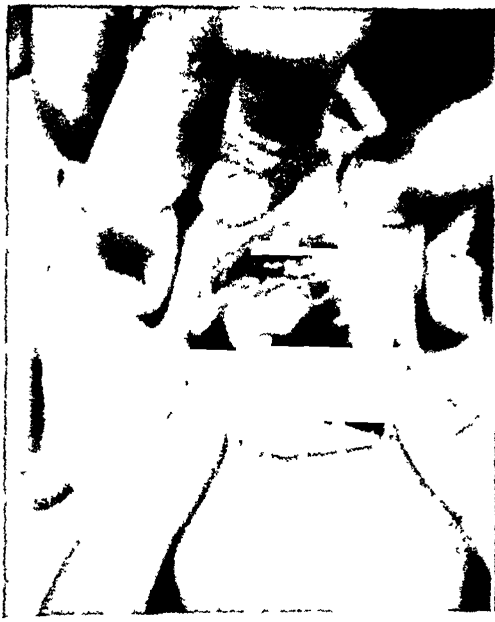

FIG 14

A case of scleroma affecting the pre-maxillary region

FIG 15.

Radiogram of case shown in Fig. 14. Note part affected.

2. The mucosal lesion gave one of the following radiological features:

(*a*) In 12 cases, the shadow was almost uniform all around the tube; only it varied in thickness sometimes at different levels (Figs. 11 and 12).

(*b*) In 2 cases, the shadow took the form of one or more tumour-like masses (Fig. 13).

These radiological features are of great value in advanced cases as they seem to be diagnostic of tracheo-scleroma when considered together with the clinical appearance of the larynx.

Affection of bone and cartilage. Scleroma is known to start in the sub-epithelial tissues and spread by direct continuity to the neighbouring soft tissues. Affection of bone and cartilage is denied (Botros, 1954). However, clinical and radiological examination of some of our cases of rhinoscleroma showed evidence of this affection and in some cases it was extensive (Figs. 14, 15 and 16). Routine biopsy material, usually taken from the superficial portion of the granuloma, does not include cartilage or bone. However, in six cases we met with, biopsy of the cartilage and bone was arranged. This material showed that this affection of the bone and cartilage was really in the nature of a pressure atrophy developing under the effect of the advancing edge of the granuloma (Fig. 17), but the granuloma itself does not grow within these structures.

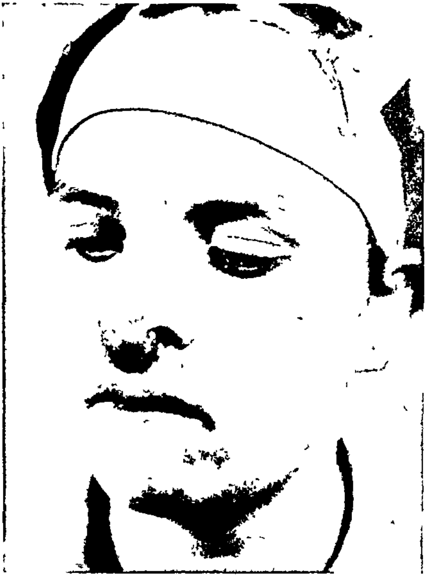

FIG 16

A case of scleroma affecting the cartilages of tip of nose

Conclusions

1. The histopathology of scleroma is reviewed. The opinion offered is that the vacuolated or foamy appearance of Mickulicz cells is due to hydropic degeneration.

2. Some clinical features not yet described or stressed have been

demonstrated. (*a*) Scleroma may take the clinical picture of a "tumour-like mass" alone or in association with other manifestations. This feature should be kept in mind in order to avoid a mistaken diagnosis of new growth or gumma. (*b*) Radiological features of tracheoscleroma are given. They are highly diagnostic in cases in which biopsy cannot be done.

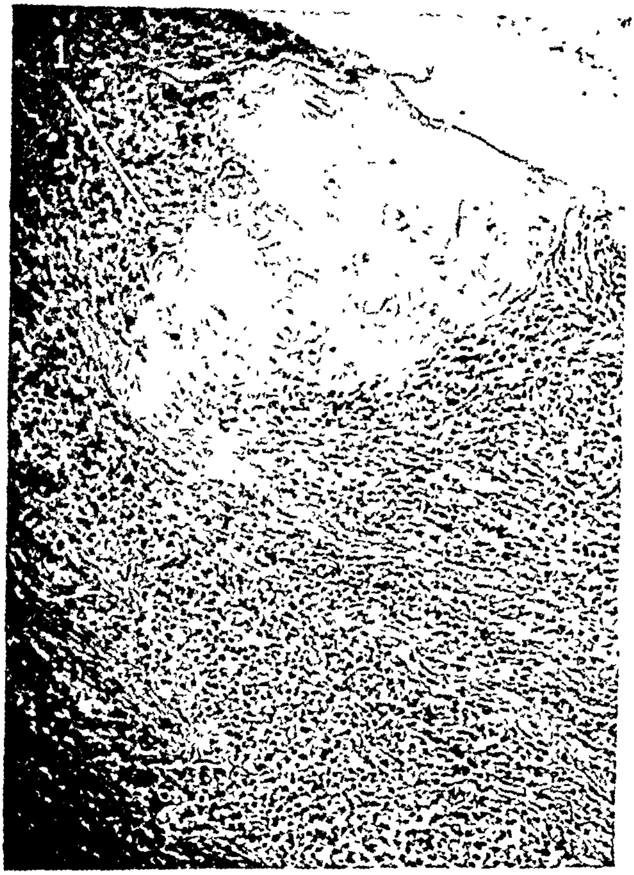

FIG. 17.

Photomicrograph of section from case shown in Fig. 16. Note the pressure atrophy of the cartilage caused by the advancing edge of the granuloma.

3. Scleroma affects bone and cartilage. Histological study of 6 cases showed that the granuloma causes pressure atrophy as it approaches them in contradistinction to its actual invasion of the soft tissues.

Acknowledgment

The authors are deeply obliged to Professor M. Hashem, Chief of the Pathology Department, for the pathological diagnosis of many of our

cases. Grateful acknowledgment is due to the Staff of the Radiology Department for their co-operation and assistance in the radiological investigations.

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ELECTRICAL NYSTAGMOGRAPHY AND ITS USE IN THE CLINICAL INVESTIGATION OF VESTIBULAR FUNCTION

By K. McLAY,* M. F. MADIGAN and F. C. ORMEROD (London)

SINCE the introduction of clinical tests of vestibular function by Bárány at the beginning of this century, many workers have sought to perfect a system of recording the nystagmus induced during such tests. Among these Dohlman (1925), in addition to demonstrating the relations between the stimulus and the speed of the slow phase of nystagmus, showed that clear, accurate nystagmus recording was possible. Since 1939 Jung and Mittermaier have developed a method of nystagmus recording which lends itself to use in clinical practice. This method, based on the electrical recording of corneo-retinal potentials projected in the skin at the lateral orbital margins, is now coming into widespread use.

Such records give details of the direction, amplitude, velocity of the slow phase, duration and character of the nystagmus, all of which are significant in the understanding of normal and abnormal vestibular function.

Henriksson (1955) has modified this technique to record directly the velocity of the slow phase of the nystagmus.

The technique and apparatus required for accurate nystagmus recording is described and an account of its use in clinical vestibular examination is given.

Method

The constant electrical D.C. potential between the cornea and the retina manifests itself at the surface of the skin at the margin of the orbit. Changes in position of the eyeball give rise to changes of this skin potential which persist for the duration of the eye deviations. These changes in potential, after amplification, are used to operate the pen of an electric pen recorder (Fig. 1). The potentials are picked up at the lateral margins of the orbits by means of silver saucer-shaped electrodes 5 mm. in diameter. Silver wires are fused to the electrodes in order to eliminate voltaic or thermo junction effects at the skin. The electrodes are firmly secured to the skin, previously cleaned with ether, by means of sellotape cellulose adhesive, and good electrical contact is maintained by the use of "Cambridge" electrode jelly A similar electrode is applied to the forehead for use as an earth lead, see Fig 2

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The orbital electrodes lead to a balanced R C coupled amplifier feeding a pen recorder The amplifier has an overall time constant of 10 seconds, the significance of which is explained as follows An amplifier with a time constant of 10 seconds will introduce an error of somewhat under 10 per cent in amplitude for a nystagmus stroke lasting 2 seconds An amplifier with a time constant of 2 seconds will introduce an error of 38 per cent in amplitude under the same circumstances

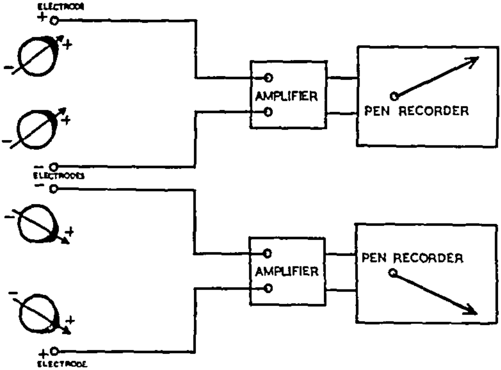

FIG 1

Method of utilizing the electrical potential changes which occur at the skin of the lateral margins of the orbits to record eye movements

The maximum useful gain of the amplifier produces a deflection of the recording pen of 1 cm for a 50 microvolt signal However, under normal operating conditions (i e with the patient's eyes closed) it is not possible to utilize the full gain of the system The average signal obtained from the orbital electrodes is in the region of 40 microvolts per degree of eye deflection The normal saccadic eye movements are about 1 degree or 2 degrees, but random eye deviations occur behind closed eyes and may well be up to 5 degrees in either direction

It has been found in practice that an amplification, which gives a pen deflection of 1 mm for each degree of eye movement, is satisfactory and that twice this amplification is useful for the determination of the end point of the duration of the course of a nystagmus

Routine Vestibular Examination

The following standardized test procedure has been adopted for the clinical examination of patients in which disorder of the vestibular system is suspected, after the usual tests for abnormalities of gait, past pointing, spontaneous nystagmus and Romberg's test have been carried out.

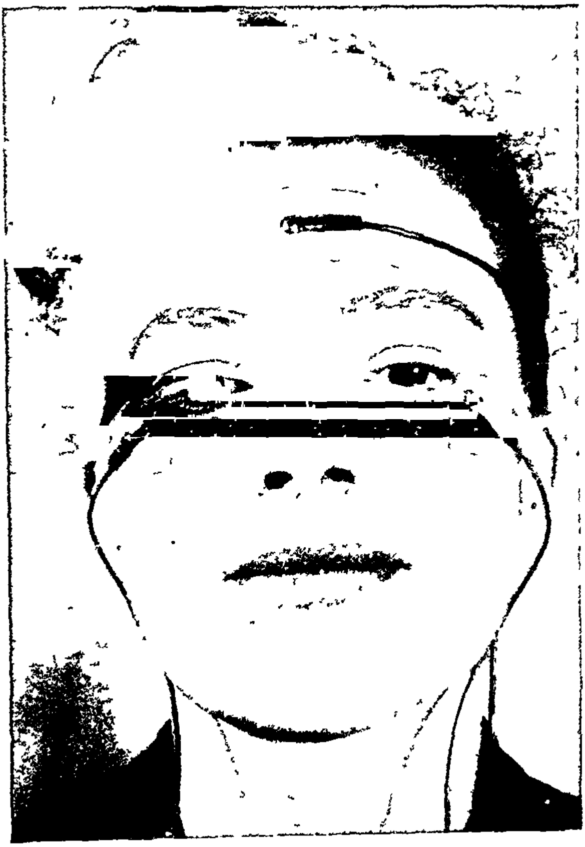

FIG 2

Electrodes in place at the outer corners of the eyes The electrode on the forehead is taken to earth

(a) *Preparation of the Patient*

The electrodes are applied to the skin at the outer margins of the orbits as already described. Calibration of the apparatus is then carried out; the patient looks alternately at two marks on the wall 20 degrees apart. This enables a suitable amplification level to be selected so that the pen deflection corresponding to an eye movement of 20 degrees is about 20 mm. (Fig. 3).

(b) Positional Test

This test is carried out to ascertain the effect of linear acceleration (in this case gravity), on the vestibular apparatus (Nylén, Bárány, etc.).

Recordings are made with the eyes closed and the patient's head in six different positions, viz: upright, supine, right lateral, left lateral, head down and prone. Angular acceleratory stimuli are excluded by this technique.

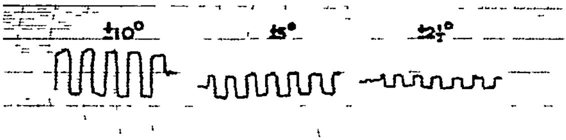

FIG 3

Calibration record, showing movements of the eyes through known angles The figures refer to movements on either side of the midline Movements towards the top of the paper represent motion of eyes to the right

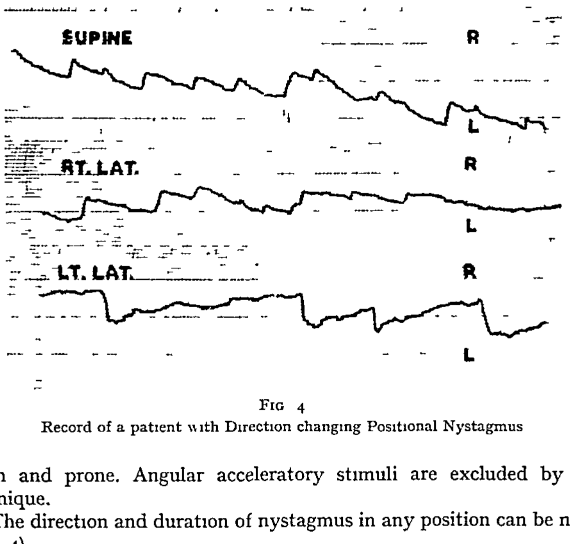

FIG 4

Record of a patient with Direction changing Positional Nystagmus

The direction and duration of nystagmus in any position can be noted (Fig. 4).

(c) Optokinetic Test

The normal following by the eye of a moving object is tested in this way and the integrity of the optokinetic pathways verified. The test is of value in the differential diagnosis of nystagmus rather than in the investigation of the vestibular apparatus but it is useful and convenient to carry out the test when nystagmography and precise electronic control of the drum speed are available.

The patient is seated with his eyes at the centre point of a drum 30 in. in diameter inside which are marked vertical stripes at 60 degree intervals. The drum is rotated at 60 degree/second for 30 seconds and the patient is asked to follow the moving stripes. He then closes his eyes when any after-nystagmus can be noted (Fig. 5).

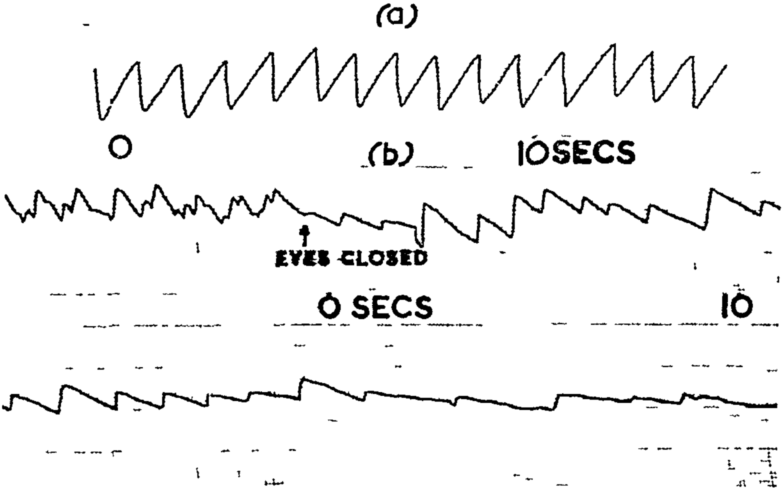

Fig. 5.

Optokinetic Nystagmus

(*a*) Normal following, with drum rotating to the right

(*b*) Sub-normal following Upon closing the eyes a well-marked after nystagmus results

Rotation Tests

Rotation tests using controlled acceleration and velocities are now well established in the investigation of vestibular function (Bárány, Tönnies, Hallpike, Van Egmond, Jongkees, etc.). The rotation stimulus is achieved by using a rotating chair, whose acceleration and velocity is controlled electronically. The stimulus used is an acceleration of 10 degrees per second^2 until 100 degrees per second velocity has been reached. The rotation of the chair at this velocity is continued while the nystagmus resulting from the acceleration is diminishing. This is the primary nystagmus. It is frequently followed by a nystagmus in the opposite direction, the secondary nystagmus. When all primary and secondary nystagmus has ceased, the chair is suddenly stopped, and the post-rotation nystagmus is recorded (see Fig. 6). This test is applied both in a clockwise and anti-clockwise direction.

Caloric Test

This test is carried out as described by Fitzgerald and Hallpike, using water at 30° C. and 44° C., except that the nystagmus is recorded with the eyes closed. Fig 7 shows a typical recording.

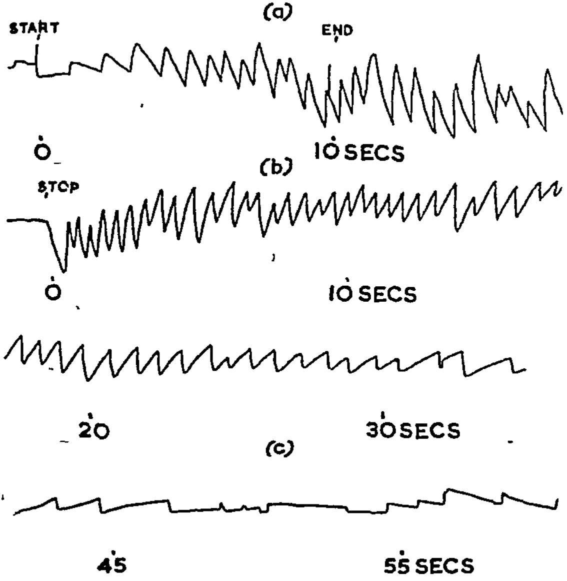

FIG 6

Rotation Test

(*a*) During acceleration nystagmus occurs with increasing speed of slow phase The speed of the slow phase is indicated by the steepness of its slope When the acceleration ends (velocity 100°/sec) the nystagmus gradually diminishes

(*b*) Nystagmus resulting when the rotating chair is suddenly stopped The nystagmus slows as the cupula returns to rest

(*c*) Primary nystagmus followed by a secondary nystagmus in the opposite direction

Discussion

Throughout the description of the method of vestibular examination, examples of typical recordings of normal subjects are given. However, in a considerable proportion of tests of normal subjects atypical records are obtained. Fig. 8 shows a record of a rotation test in a normal subject, where the normal nystagmus reflex is much reduced. This is one of the many subjects where the reaction was reduced or absent.

In many normal patients, secondary nystagmus can be recorded. In the interval between the primary nystagmus and the secondary nystagmus there is frequently evidence of quick phase eye movements in each direction, as if nystagmus in each direction were present at the same time. These movements when recorded form a typical pattern which can be conveniently described as Square Waves. This phase of the nystagmus reaction is sufficiently consistent to be of value in accurately assessing the end point of a primary nystagmus (see Fig. 6(*c*)).

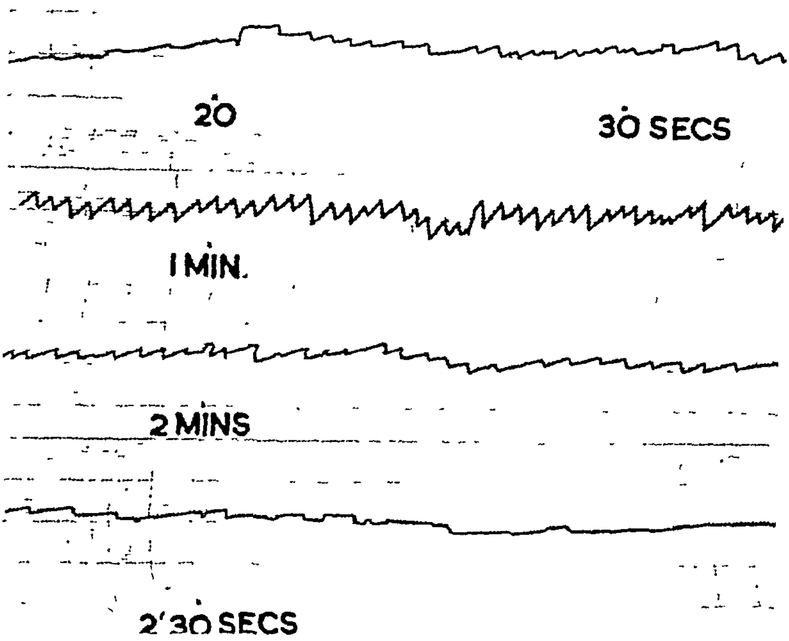

Fig. 7.

Recording of nystagmus during douching of the right ear with water at 30° C. for 40 sec.

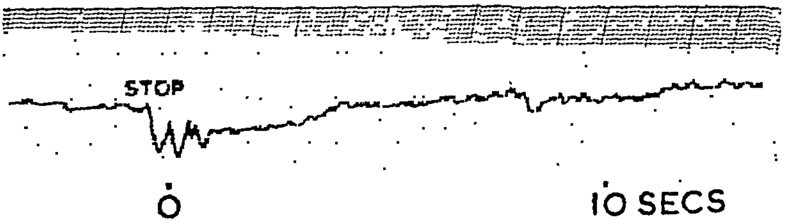

Fig. 8.

Nystagmus on cessation of rotation inhibited after two or three strokes.

This type of eye movement has also been frequently found to occur spontaneously, when recordings are made behind closed eyes, in cases of diagnosed vestibular neuronitis (see Fig. 9).

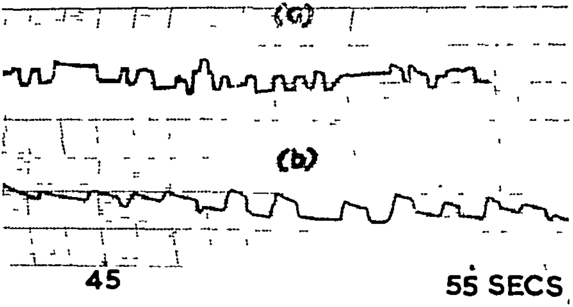

Fig 9

Two different patients, diagnosed as vestibular neuronitis, where square waves occurred spontaneously

Conclusion

At present, by the method described, much reliable information can be obtained about eye movements occurring in response to vestibular stimulation From such records the presence of nystagmus can be detected, its duration, and the speed of the slow phase obtained. In addition it seems probable that further experience in this method will demonstrate the importance of forms of eye movement other than nystagmus in the diagnosis of vestibular abnormalities

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MYRINGOPLASTY

THE SURGICAL REPAIR OF TYMPANIC MEMBRANE PERFORATIONS

By S. R. MAWSON (London)

A DEFECT in the tympanic membrane has always been a provocation to otologists. It is an æsthetic imperfection that offends the eye, an exasperating impediment to function that reproaches final acceptance, and it is an open invitation to further infection and robbery of hearing that begs for the simple insurance of closure.

The multiplicity of ingenious methods devised to meet this problem indicates the inherent difficulty of solution. The defect may be approached in one of three ways. It may be plugged; it may be patched; or it may be stimulated to close itself.

Plugs

The simplest plug is the piece of cotton wool soaked in sterile paraffin, which is used to occlude the perforation and contact the medial wall of the middle ear. It still has its uses and often may be successfully positioned by the patient. The most effective plug is probably a hollow cone, or conical cylinder, of inert plastic inserted through the perforation to make contact, as with the cotton wool, with the medial wall of the middle ear (Pohlman, 1951). However, such an artificial membrane needs constant attention as it is apt to cause recurrences of otitis media and otitis externa and does not obviate the necessity to remain on "precautions".

Patches

The patches recommended in the past have included rubber, sized paper, elastic, collodion, egg membrane, sheep's membrane, cigarette paper, gold foil, and, most recently, human amniotic membrane (Schrimpf, 1954), which would appear in principle and practice to be the most successful.

Stimulators

Of the agents used to stimulate the margins of a perforation to proliferate, so as to achieve ideal natural repair, trichlor-acetic-acid has proved the most active. Repeated applications are sometimes attended by success. But results are unpredictable, treatment is tedious and the method is only suitable for the smaller perforations.

Human Amniotic Membrane

It was dissatisfaction with these methods which led Schrimpf (1954) to try specially prepared human amniotic membrane. Out of 53 perforations successful closure was obtained in 48, in an average of 43 days. Three failures were due to infection and two could not be explained. Average improvement in hearing in the successful cases was 18 db (air conduction). These results are extremely good but it should be observed that the length of follow-up is not disclosed and that the method is only suitable for central perforations where there is a margin of healthy membrane surrounding the perforation.

Present Method

It was the stimulus given to the study of this subject by Schrimpf, together with the possibilities of skin grafting as demonstrated in the tympanoplastic operations of Zöllner (1954) and Wullstein (1955) that renewed the desire to search for a *standard* surgical procedure that could be applied simply, shortly, and effectively to closure of a perforation. *Especially to the large, sub-total perforation which generally evaded* other methods. It is desirable to emphasize the word "standard". So much reconstructive surgery of the middle ear has recently been in a state of flux and experiment, with the final surgical disposition left to the devices of the individual operator, that it seemed it might be a helpful step to look for a simple standard procedure for this one aspect of the middle-ear problem; namely, large, dry, sub-total perforations.

Principles of Skin Grafting

The first step was to re-examine the general principles underlying skin grafting. These are dealt with very adequately by Matthews (1943) in his book, *The Surgery of Repair*, and I am indebted to him for the information I was able to obtain therefrom.

The overriding problem is one of nourishment. The *pedicle graft* safeguards blood supply by allowing a new source to be established before the old is severed. I have made one or two attempts to fashion a pedicle graft from the skin of the meatus but a promising theoretical approach has in each case proved a failure. Others, notably Frenckner (1955), appear to have met with more success.

Turning to free skin graft, these are of three main types.

(1) *Pinch grafts*

(*a*) Thin (Reverdin, 1869). Circular pieces of skin laid in rows 1 cm. from each other.

(*b*) Thick (Davis, 1914). This is essentially a Reverdin modified to a full thickness centre with shelving edges. Such a graft is easy to cut, is hardy, survives in sepsis, but on the other hand scars the donor area, gives indifferent recipient cosmesis and is largely used as first aid grafts over tendons and joints to preserve movement.

(2) *Razor Grafts*

(*a*) Thin (Ollier, 1872; Thiersch, 1886). Consists of superficial layers of epithelium, differing only in size from Reverdin. Useful on any raw surfaces, except muscles. May be applied to granulations but liable to contract by one quarter.

(*b*) Thick (Blair, 1929). Split skin grafts with Blair or Humbey knife are whiter, more opaque than thin grafts through which blade is distinctly visible when cutting. Has the widest application of all free grafts. Moderate pliability. Even texture. Good cosmesis. Strong enough to withstand pressure. Survives minor sepsis.

(3) *Full Thickness Grafts*

Wolfe (1867), Krause (1893). Advantages are of cosmesis, pliability, ability to withstand pressure but are more delicate than razor grafts and will not survive in sepsis. Always perish over a hæmatoma. Inclusion of fat results in necrosis. Useless for granulating areas. Full thickness grafts depend on absorption of lymph from their healed surfaces for nourishment and therefore are made to fit the recipient exactly. (Razor grafts being thinner can absorb nourishment from a small base and can be cut large to allow for contraction.) It is advocated by Matthews (1943) that the Wolfe-Krause graft is left undisturbed for 7 days and it will almost always be found to desquamate.

(4) *Survival of Graft*

From studies made of grafts at intervals after being laid on their bed, Wright (1956) concludes that the first two or three days of the graft's survival are dependent on intra-vascular graft circulation. This is especially based on the fact that four hours after placing of the graft the intrinsic vessels of the graft were observed to be in spasm but 24 hours later these vessels were relaxed and full of red blood corpuscles. (Others, notably Conway (1951), feel that much of the early survival is due to extra-cellular nourishment by imbibition.) From 16 hours onwards there is proliferation of endothelial blood from the host's blood vessels. At 72 hours continuity of host's and graft's blood vessels has been noted and normal circulation has been seen established by the eighth day. Preliminary fixation of the graft occurs within 5 hours by a fibrin clot. This gradually retracts and is invaded by leucocytes and phagocytes. On the 4th and 5th day fibroblasts begin to invade the clot and organization is fairly complete by the 10th day.

Selection of Graft

Experience has shown that the razor grafts are very useful in mastoid cavities and it might be expected on theoretical grounds that this usefulness would extend to the repair of tympanic membrane perforations. However, one of the great drawbacks is difficulty in technique. To insert a razor graft down a narrow meatus so that it lies exactly in position, without the edges curling and the surface wrinkling, is not easy and in my experience it has not lent itself well to the purpose, nor, in the few cases I have attempted, survived long or resulted in repair.

Full thickness grafts (Wolfe-Krause) on the other hand have already been used successfully in tympanoplasties and in a report of 350 cases Wullstein (1955) includes 89 of simple myringoplasty, in which the results are said to be better as regards hearing than might be anticipated from fenestration in cases of otosclerosis with similar preoperative hearing loss. Such large numbers overshadow the present series but again it is emphasized the main purpose has been to try to evolve a standard procedure from what has been, and still is, largely experimental work. Full thickness grafts are pliable and have a greater ability to stand pressure than razor grafts but they are said not to survive in sepsis and always to perish over a hæmatoma (Matthews, 1943). Moreover, inclusion of fat results in necrosis and they cannot be expected to survive on granulation tissue.

Selection of Suitable Cases

The second step was to study the factors relevant to the successful application of a full thickness graft to the defective membrane. As previously stated, the large, sub-total perforation was the one that suggested itself, since there was no really satisfactory method of dealing with it. In view of the limitations of the full thickness graft, described above, it was decided that no case of active sepsis would be considered and no case would be operated on unless the ear had been dry for at least two months. Since vigorous blood supply seemed essential for the establishment of the graft and yet hæmatoma formation under the graft would cause it to perish, a preliminary stripping of superficial epithelium from the tympanic membrane remnant and from the inner third of the external auditory meatus to form a suitable bed, was considered desirable. Furthermore, that the graft, once placed, should be maintained in position with gentle pressure to discourage hæmatoma formation. Satisfactory stripping of the membrane indicated the use of an operating microscope. While the use of a preliminary vaso-constricting injection, as in the Rosen operation, would reduce the difficulties arising from the field being too much obscured by blood.

First Objective

The first objective of the operation is to restore hearing, but the degree of deafness, especially with perforations, is very variable and bears a less close relation to the size, shape and position of the defect than might be imagined. For natural maximum amplification to occur *viâ* the conducting mechanism the membrane must be intact, the ossicular chain mobile, the eustachian tube patent and the cochlear potential must be unimpaired. With rare exceptions a perforation is a legacy of otitis media and resultant deafness depends not only on the defect itself but also on any other damage the conducting mechanism may have sustained.

In the process of healing from otitis media, adhesions and false membrane formation is common; partitioning of the tympanum with fortuitous air-tight loculations may result in pseudo-healing and good function in the presence of a large and obvious defect in the membrane. On the other hand, as is well known, the ossicular chain may have become fixed while the membrane has healed over, giving a surprising disparity between the otoscopic appearance and the measureable function.

Thus restoration of hearing is a more complex problem than the mere successful occlusion of a perforation and prognosis as regards the benefit to the hearing after closure must be guarded.

Second Objective

Protection of the middle ear.

The protection gained from closure of a perforation is a much more certain result of the operation than improvement in hearing and even if preliminary tests would suggest, due perhaps to blocked eustachian tube or fixed ossicles, that the hearing gain will not be significant, there still remains the benefit of a closed middle ear, which in itself is a gain to the patient. If an intact membrane is secured, and it will be shown that this is predictable in a high proportion of cases, a patient hitherto on "precautions" may once again go swimming and indulge in shower-baths and shampoos with impunity. Moreover, the risk of reinfection and "flare-up" will be materially reduced and prognosis for insurance and similar purposes greatly improved.

Third Objective

The standard procedure to be described has been pursued not only with these first two objectives in mind, but also with the final objective of a quick and reliable procedure causing minimal disturbance to the patient. For this reason cases with a narrow meatus have been excluded from this series. It seemed desirable to limit external skin incisions solely to that required for the obtaining of the graft and to avoid incisions in and around the meatus which prolong the post-operative period and are a source of discomfort and possible infective troubles. The simpler the procedure the shorter the stay in hospital, and this in itself seemed to merit consideration.

The Operation

Stage I—Cutting the Graft

An area on the posterior aspect of the auricle and mastoid of the ear to be grafted is marked out with ink.

A sub-epidermal purse-string catgut suture, a modification of that advocated by Björk (1955), is then placed to facilitate later handling of the graft.

With the small Bard-Parker blade the area previously marked out is incised and the skin required dissected off. The graft is then placed in normal saline, body temperature, until required. It has been suggested that placing of grafts in normal saline is injurious and prejudicial to their survival. This has not been my experience. The defect left by removal of the graft is closed after under-cutting the skin in all directions. As a matter of experience I have found that a more satisfactory scar results if this defect is repaired from below upwards. Any redundant skin thus tends to collect behind the top of the ear where it is less noticeable. The wound is closed without drainage.

Stage II—Preparation of the Receptor Area

Under magnification of 10 or 16 times with a microscope, in this series a Zeiss, a view may generally be obtained of the middle-ear region through an aural speculum, which permits detailed study and preparation. A circular incision is made round the meatus at the junction of the inner and middle thirds. The skin is dissected inwards towards the tympanic ring, (if it is dissected outwards from the perforation it will tend to enlarge the perforation as Beales has pointed out), and, in the case of a total, or subtotal, perforation, is retained and swung inwards, thus partially occluding the defect with a raw surface facing outwards. Where there is a significant amount of tympanic membrane still remaining the skin from the meatus is excised and the superficial epithelium from the tympanic membrane is dissected and swung inwards instead. If after this a large unsupported area still remains, the middle ear is filled with gelatin sponge soaked in penicillin to provide a scaffold for the graft. This preparation is achieved with instruments devised by Zöllner and the whole procedure is slow, finicky and rendered difficult by bleeding.

Stage III—The Placing of the Graft

When it has been judged that the preparation of the area is complete, that a sufficiently raw area with free blood supply has been obtained to supply adequate nourishment for the free graft, the latter is taken from the saline and stretched over the forefinger, where, with small fine scissors, all superficial fat is removed.

A small, cylindrically-shaped piece, cut from a plastic sponge, is then placed with forceps into the heart of the outer surface of the graft and the purse-string catgut suture tightened round it, the raw area of the graft thus being everted into the desirable shape for the bed prepared.

Thus controlled, the graft is inserted steadily down the meatus until it comes to lie in the desired position. The sponge is held firmly in position while the catgut suture is withdrawn by steady traction. The meatus is finally dressed with another piece of plastic sponge and an outer cotton-wool tampon. A pressure dressing is useful to prevent accumulation of blood in the donor area.

General Points

It is essential to remove the hair for at least 1½ in. from the area from where the graft is to be taken, and this area should receive overnight preparation to ensure sterility. The meatus and middle ear may be conveniently cleansed with spirit at the time of operation. The operations have been performed under cover of a long-acting penicillin preparation, bacterio-static blood levels being maintained for one week post-operatively.

The stitches are removed on about the fifth day, at which time it may be found convenient to remove the sponges. The sponges having been previously moistened with sterile paraffin generally remove easily but if any difficulty is encountered in extracting the deep sponge it is advocated that this is left for 2 or 3 weeks to ensure that the graft is established and will not be brought out with the sponge. Any discomfort or discharge, or other complication, may necessitate removal of the sponge and possible loss of the graft.

The operation occasions minimal disturbance to the patient and full activity may be resumed on the second post-operative day. The post-auricular wound heals quickly, if only with the slight defect of a more retracted auricle than previously. With care, the suture line may be made to occupy the natural post-auricular sulcus.

The only complication so far encountered has been a temporary paralysis of the VIIth nerve, ascribed to a pressure axonotmesis, which recovered completely after immediate removal of the sponges.

Myringoplasty

RESULTS

1. *Closure of Perforation*

TABLE I.

Myringoplasty	No	Intact	Perforated
Ears operated	21	0	21
Followed up for 1 month ..	21	21	0
Followed up for 4 months ..	17	16	1*
Followed up for 8 months ..	10	10	0
Followed up for 12 months ..	7	7	0
Followed up for 18 months .	5	4	1*

* Acute otitis media

Nineteen cases out of 21 were closed

2. *Hearing*

(*a*) *Patients' Estimate*

TABLE II

Myringoplasty	No	Improved		No change	Worse
		Marked	Slight		
Followed up at 1 month	4	2	1	1	0
Followed up at 4 months	7	1	5	1	0
Followed up at 8 months	3	0	1	2	0
Followed up at 12 months	2	1	1	0	0
Followed up at 18 months	5	2	1	2	0
Totals	21	6	9	6	0

Fifteen cases out of 21 were improved (two-thirds)

Six cases out of 21 were markedly improved (one-third)

(*b*) *Audiometric Estimate*

TABLE III

AVERAGE AUDIOGRAM OF 6 CASES WITH MARKED SUBJECTIVE IMPROVEMENT

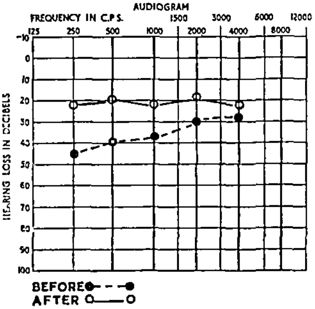


TABLE IV.

AVERAGE AUDIOGRAM OF 9 CASES WITH SLIGHT SUBJECTIVE IMPROVEMENT.

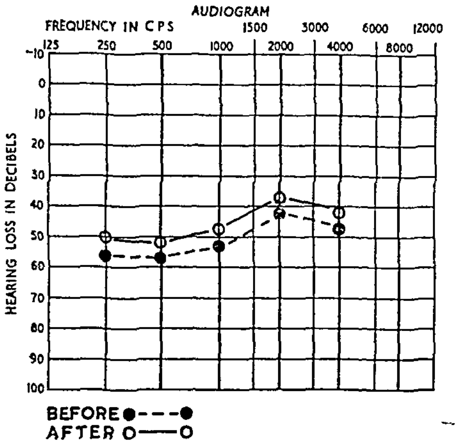
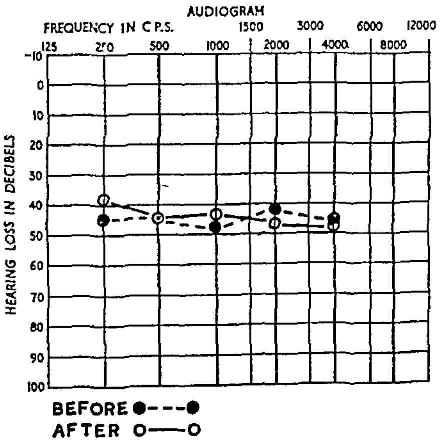


TABLE V.

AVERAGE AUDIOGRAM OF 6 CASES WITH NO SUBJECTIVE CHANGE.



3. *Comparison with Other Published Results*

TABLE VI.

Author	Operations	Takes	Hearing		
			Improved	Unchanged	Worse
Wright (1956)	56	40	25*	5	0
Beales (1957)	17	16	12	5	1
Mawson (1957)	21	21	15	6	0

* Ten cases untested.

Discussion

From the results of this operation and of similar operations in other hands, it would now seem established beyond doubt that surgical reconstruction of tympanic membranes is a practical proposition Using full-thickness skin grafts, denuded of superficial fat, and placed in a healthy, microscopically prepared bed, a high proportion of "takes" can be anticipated In this series immediate "takes" were 100 per cent and out of 21 cases only 2 subsequent perforations occurred, due in each case to otitis media

The results as regards hearing are less predictable but nevertheless still favourable, in that there seems to be a two-thirds chance of improving the hearing and a one-third chance of obtaining really marked improvement The pre-operative level of hearing has an obvious bearing on the prospects of improvement Losses that do not exceed 40 db anticipate the best results

The value of special pre operative tests with such instruments as the acoustic probe has been considered but in view of the fact that a high proportion of permanent closures may be hoped for and that the possibility of the hearing being made worse is extremely remote, it was concluded that no real value attaches to these tests An adverse test, i e one suggesting ossicular discontinuity, would not be a deterrent to recommending operation in order to provide the protective advantage of closure Furthermore, it was observed that marked improvement of the order of 25 db occurred in three cases where the starting point in some frequencies was 50 db, a level often said to be associated with ossicle chain rupture The re-statement may bear repetition that the main object of this work has been to attempt to establish a sound basic procedure with as few variables as possible The operation described is not considered to be more than a platform from which to launch further attacks on the problem of restoration of function after middle-ear disease It is hoped that many improvements will have already suggested themselves The method of inverting the outer layer of the tympanic membrane or meatal wall into the perforation, is considered an improvement on the use of gelatin sponge or scarification of the promontory The concept of preparing a large bed for the graft, and merely cutting the latter to ensure that it is ultimately of a slightly smaller surface area than the bed, may seem clumsy but it has proved itself in practice I feel I may conclude by saying three patients have requested that their other ear be done and one was so pleased about it that he actually wrote to the Minister of Health to tell him so

Summary

Defects in the tympanic membrane have always been a provocative challenge to treatment, the multiplicity of ingenious methods evolved

indicating the inherent difficulty of solution. Previous treatments having failed to prove entirely satisfactory, recent attention has turned to skin-grafting, the principles of which are reviewed. An operation for the surgical reconstruction of the tympanic membrane is described in detail, depending for success on strict asepsis and microscopical preparation of the receptor site. Analysis of results shows that a high proportion of graft survivals can be anticipated. The results as regards hearing are less predictable but indicate a two-thirds chance of improving the hearing and a one-third chance of obtaining really marked improvement.

It is concluded that this simple operation represents one platform from which to launch further attacks on the problem of reconstructive surgery of the middle ear.

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ON TYMPANOPLASTY

By NILS G. RICHTNÉR (Stockholm)

PLASTIC surgery of the middle ear has gained tremendous ground in otology, especially through the outstanding work of Professors Wullstein and Zöllner. A patient with a chronic middle-ear inflammation, frequently with greatly impaired hearing, may now be relieved of his infection and the false tumours (so-called cholesteatomas) frequently accompanying these forms of ear inflammation as well as having his hearing improved to a certain extent. Thanks to the advances made by these eminent workers in otological surgery this has become possible not only as earlier through radical operations on the cellular system of the ear but also now through plastic operations on the elements of the middle ear, the mucosa, the auditory ossicles and the tympanum, and in many cases with astonishing improvement to the hearing. There are primarily five different types of operative approach involved.

The plastic operations on the middle ear are included under the common name of tympanoplasty. These all involve a radical approach, either peraurally or retro-auricularly, with operation on the cell system of the ear and creation of different types of middle-ear cavities. These operations are suitable for all forms of chronic middle-ear inflammation and may be performed in a single stage operation or, if the inflammation is very extensive and acute, in two stages. In these latter cases radical operation is first done on the cell system of the ear with elimination of the inflammation and the cholesteatoma that may be present, and a few months later, in a second step, when the middle ear and the radical cavity have healed, the actual tympanoplasty is performed.

All these operations are preceded by a thorough clinical study of the middle ear, frequently under magnification, for evaluation of the appearance and type of the perforation and investigation of the auditory ossicles. With the co-operation of our audiologist special measurements of the hearing are always carried out, primarily with the ordinary pure tone audiogram and sometimes complemented with a speech audiogram. Our audiologist, Lundborg, also assisted the investigations with sonometric studies following Zöllner's pattern. At Söder Hospital these studies are called transmission investigations and include, in addition to the actual vibrating probe, prosthesis tests—both so-called complete prosthesis tests and covering prosthesis tests. The latter involve primarily screening prosthesis over the round window. The methods of transmission investigation have been developed to such an extent in the Department of Otology at Söder Hospital that we are able, by this means, even before the operation to a comparatively great extent, to obtain a conception of the condition of the ossicle chain and what screening of the round window will be able to afford in the way of improvement in hearing. Therefore we are able, in a large percentage of the cases, to determine preoperatively the type of operation to be preferred. In many cases this is of tremendous value. The clinical investigation also naturally provides us with a preconception of possible measures and of the type of operation that should be done, but the transmission investigation expediently complements these clinical preliminary examinations and would seem to give this form of surgery a broader scientific foundation.

The different forms of tympanoplasty we perform agree, on the whole, with those of the German otologists.

In chronic middle-ear inflammations, where we have chronic mastoiditis or cholesteatomas of lesser extent that can be removed without injuring the auditory ossicles or tympanic membrane and where the ossicles are in good condition with contact existing between all ossicles, after conservative radical operation a plastic graft is placed over the entire tympanum and the operative cavity, forming a so-called Cavum major cavity according to German nomenclature "eine grosse Pauke". In a case presenting minor destruction of only the long process of the incus we try transposition of the incus so that its long process comes into contact with the stapes.

Transposition of a carious incus so that contact is obtained has been done in four cases. In one the results were particularly good. In these cases a cavum major was constructed despite a defective ossicle chain. This patient has an improvement of more than 40 db in her hearing. The auditory improvement is computed according to accepted methods on the basis of the values obtained in the audiogram for frequencies of 500, 1,000, and 2,000 c.p.s., taking the mean value for these three. Before the operation the mean loss of hearing was 78 db, and now, one and a half years after, it is 35 db.

As a rule with the cavum major procedure a good auditory result is obtained, frequently with values lying about 10 to 20 db. Nor is this particularly surprising, inasmuch as the auditory ossicles are in good condition before the operation. It is merely a matter of performing the operation as smoothly and gently as possible, retaining as much undamaged epithelium as can be salvaged and being careful not to create an opportunity for adhesion formation post-operatively. Among other precautions to be observed, it is necessary to be particularly careful not to injure the extremely thin mucosal epithelium on the auditory ossicles. I am thinking especially here of the incus where the mucosa is easily damaged if special care is not taken to avoid trauma. I also retain as much as possible of the mucous membrane of the antrum both on the floor over the semicircular canals and over the incus. An incision is made so the mucous membrane may be raised to permit inspection of the auditory ossicles. On the other hand, it does not seem to matter that we free the tympanum at the limbus and lift it forward to allow inspection of the middle ear. This manipulation does not appear to damage the hearing. The mucosa of the middle ear is disturbed as little as possible. Granulations are carefully lifted out or snared. Curettes must not be used. Introduction of foreign bodies such as oxycel or spongostan into the middle ear is to be avoided if possible because of the risk that they may not be absorbed and may have a deleterious effect on the auditory results. With this procedure we dare to tackle even cases of chronic otitis with chronic mastoiditis and an auditory impairment of from 20 to 25 db.

Type 3 (Wullstein) of the plastic middle-ear operations is the co-called tympanostapediopexy or according to German nomenclature, the formation of "eine flache Pauke". This is used as a rule in cases where there is pronounced destruction of the ossicle chain, but a relatively intact stapes The residual tympanum is then dropped with the accompanying plastic flap down against the head of the stapes, thereby achieving transmission of the oscillations directly from the tympanum to the stapes. We do not usually use a columella. I make it a rule not to introduce foreign bodies into the middle ear if it can be avoided. The primary results of this type of operation have been good both with respect to healing and hearing, but the late results have been poorer in some cases, frequently due to the occurrence of a perforation in the flap over the stapes.

Instead, in the cases where we have a pronounced defect in the ossicle chain, I have employed type 4, the so-called cavum minor technique. In many cases this has afforded astonishingly good results even in cases of extremely grave auditory impairment. Many encouraging results were obtained with this technique, even in the early days two years ago when I started seriously to engage in tympanoplastic operations. Stimulated by the excellent results, I have therefore been particularly interested in undertaking treatment of the extremely grave cases of auditory impairment in chronic otitis in order to see what results can be attained with this special technique. Among about 100 tympanoplastic operations I have performed at Söder Hospital no less than 39 patients were operated on according to the cavum minor technique, with minor variations from case to case, but with the consistent purpose of establishing a difference in pressure between the fenestra ovalis and the fenestra rotunda. It will consequently be this type of operation I shall discuss in detail now, particularly with respect to technique, diagnosis and primarily the auditory results.

As in all other plastic operations on the middle ear, it is also true in this case that the tube should be patent, permitting easy passage of inflated air. The operative procedure is largely as described by Wullstein and Zöllner. First the radical operation is performed with thorough removal of polyps and granulations about the residual stapes or the footplate of the stapes and the round window and tube without damaging the epithelium more than necessary. An incision is then made in the mucous membrane of the medial wall of the middle ear below the prominence of the facial nerve canal and farthest forward over the tube above the facial prominence so that a narrow flap is obtained there to fold up against the tube. The rest of the mucous membrane is turned downward. The epithelium forms a bed for the transplant. At operation it is generally found that part of the limbus is still present. This shall be treated with great care as it provides an excellent bed for the transplant, which is obtained retroauricularly in all cases in conjunction with the application of the retroauricular incision. It is important that there can be an open communication between the hypotympanon, the tube, and the round window. The transplant is subsequently held in position with oxycel, which is treated with penicillin, hyaluronidase and prednisolone trimethylacetate (Ultracortenol, Ciba). As a rule this is absorbed within a week or 10 days. The fenestra ovalis is exposed, with or without the stapes. Another full skin graft is applied over the defect remaining in the mastoid process and the upper part of the middle ear.

The cases we selected for this technique comprised chronic adhesive otitis media—adhesive otitis with a healed thin scar adherent to the promontory or with a small perforation in the thin scar with bad hearing. Roentgen examination has shown chronic mastoiditis. Five such cases were operatively treated. Seven cases of chronic otitis media with attic or antrum fistula with cholesteatoma are included in the series. In addition there are five cases of chronic otitis media with attic or antrum fistula without cholesteatoma. There are other groups with chronic otitis with central perforation and chronic mastoiditis. Five such cases including one with dry ear and four with suppurating ears were operatively treated. The series also includes 11 cases of earlier radically operated chronic otitis with continued discharge and greatly impaired hearing as well as six cases of clean radically operated cavities with greatly impaired hearing. Altogether the series comprises 39 cases.

A somewhat more detailed review of the condition of the auditory ossicles and the mucous membrane of the middle ear observed at operation in these cases would perhaps be of value here. Special attention will be given to the importance of a stapes in good condition as opposed to a residual stapes base only. It will also be possible to observe the auditory results when the operation revealed granulation, free fenestra ovalis and rotunda.

This series includes three cases in which the malleus and incus were found at operation to be absent. Only the stapes, which was movable, could be seen. In seven cases only a stapes base remained. In 19 cases the ossicles were extremely carious, and in 10 cases the malleus, incus and stapes, except for the base, were removed. In nine cases the stapes, which had good mobility, was retained.

In eight cases the ossicles were in rather good condition, but caries was present and no contact existed between the incus and stapes. In three cases the malleus was left, and in five both the malleus and incus. The stapes and fenestra rotunda were granulation free in three cases, while there were abundant granulations in 27 cases in the regions of both fenestrae. The mucous membrane of middle ear was otherwise clear in nine cases but granulated in 26. No granulations in the tube were present in 30 cases, but granulations were observed in the tubal orifice in seven.

Description of Some Cases and Auditory Results

One case is worth detailed description. This patient's hearing became very good, almost unbelievably good, and certainly far better than could have been expected. It is the case of a 25-year-old woman who had earlier undergone radical operation with removal of osteitic malleus and incus. The radical cavity was clean and clear in this patient, but her hearing was poor, with 51 db the mean level for 500, 1,000, and 2,000 c.p.s. The tympanum presented a large perforation covering about three-quarters of it. The stapes was freely visible and easily accessible for vibrating probe, which showed good transmission *viâ* the stapes. Revision of the radical operation was done with tympanoplasty with a free transplant (cavum minor). The stapes lay embedded in granulations but was satisfactorily movable. The niche of the fenestra rotunda was very deep, without granulations. The tube was normal and the middle ear mucosa otherwise pale and normal. The mucous membrane was divided in the usual manner on the promontory and folded downward. The mucosa was removed from the epitympanum, and a transplant was applied there. Thereafter a cavum minor was created in the usual manner. The ear healed rapidly. After only one month the auditory level was 33 db and now, after 18 months, is not only still good but has shown further improvement to a mean of 21 db from an initial preoperative 51 db. Accordingly, there has been a thus far permanent improvement of 30 db. The stapes is now covered by a thin epithelium, and the fenestra rotunda is well isolated under the plastic flap. The tube affords air passage. This is another case in which it is difficult to explain the excellent resultant hearing. The patient has only the stapes left and has screening of the fenestra rotunda. She now hears a whisper at 1 m. and conversational tone at more than 5 m. with the operated ear. The auditory improvement in db is shown in Fig. 1.

One more case in this series will be reported as it demonstrates how it is possible for an auditory invalid to recover good hearing through tympanoplasty and to return to his clerical position without needing to use a hearing aid. Both ears had earlier been radically operated on for cholesteatoma. In the right ear there was a previously created fistula over the horizontal semicircular canal, and the fenestra rotunda had been plugged with resultant exceedingly poor auditory effect. In addition there was an auditory impairment corresponding to a mean of 71 db. The right ear perceived conversation at a distance of 0·4 m. and the left of 1·5 m. Revision of the operation on the left ear was done with creation of a cavum minor. All auditory ossicles were gone with the exception of the footplate of the stapes, which was movable. There were granulations over the fenestra rotunda and ovalis and granulating middle-ear mucosa as well. The tube was fully patent. An optimal flap was obtained over the hypotympanum. After only 14 days there was appreciable improvement in hearing with an increase from an initial level of 71 db to 53 db. After another 14 days the improvement had progressed to 43 db, and the patient perceived conversation at a distance of more than 5 m. His hearing has since consistently been very good, now being at a level of 41 db 18 months after the operation. He is able to hear conversation up to a distance of 15 m. In this case only the base of the stapes remained and no new columella was introduced. Because of the extensive polypous

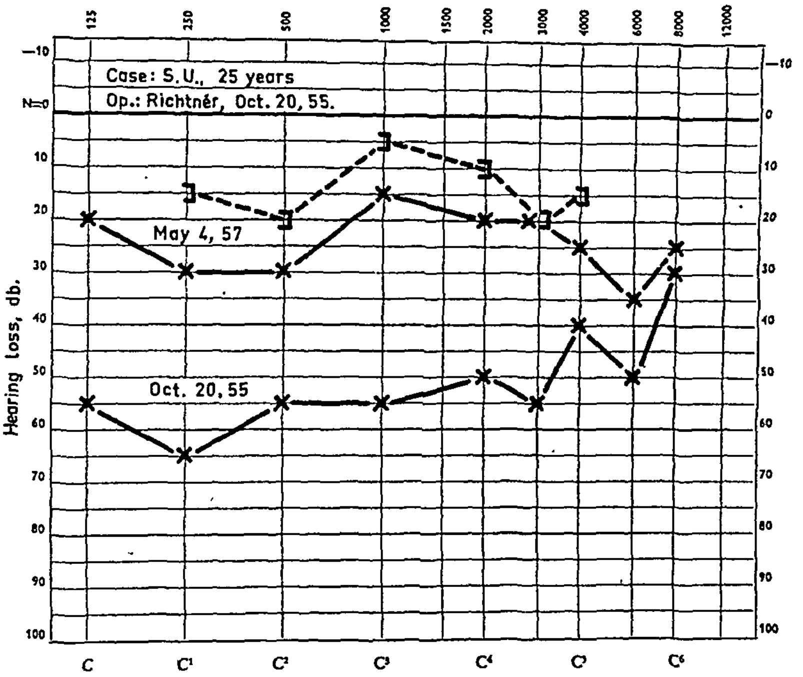

FIG. 1.

granulations in the middle ear much of the mucous membrane had to be removed Therefore oxycel was applied as a bed for the transplant in order to allow time for the mucosal epithelium to develop and to prevent the formation of adhesions between the flap and the promontory In this case the use of oxycel in the middle ear proved advantageous rather than the contrary However, this should not be taken as grounds for generalization, and introduction of foreign bodies into the middle ear should be avoided whenever possible If anything is to be used in such cases as this,

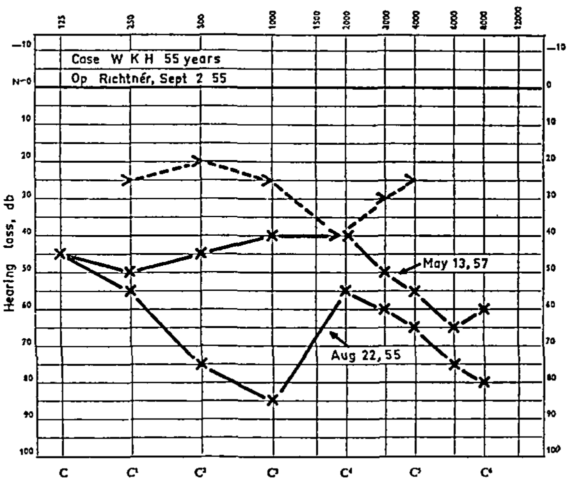

FIG 2

it is my experience that oxycel saturated with penicillin, hyaluronidase and ultracortinol is best, but only applied in small bits and not in large clumps (see Fig 2)

Another case was that of a 30-year old man who had undergone radical operation some years earlier for cholesteatoma The incus was removed at that time, but the malleus was left *in situ* There was considerable loss of hearing, and the ear did not heal, suppurating from time to time Bilateral tubal catheterization revealed nothing abnormal Revision of the radical operation was done with tympanoplasty and free transplant (cavum minor) A cholesteatoma was encountered at the site of the head of the malleus No residue of the malleus was found The limbus of the tympanic membrane was still partially present There were abundant granulations in the region of the fenestrae ovalis and rotunda. The stapes was easily movable. The tube was fully patent and without granulations. Cavum minor was created in the usual manner. The upper part of the middle ear above the fenestra ovalis was denuded of mucous membrane, and the transplant was applied directly to the bone there. An ideal flap was obtained from above the hypotympanum. Fourteen days after the operation there was considerable auditory improvement. The improvement continued subsequently, and now, six months after the operation,

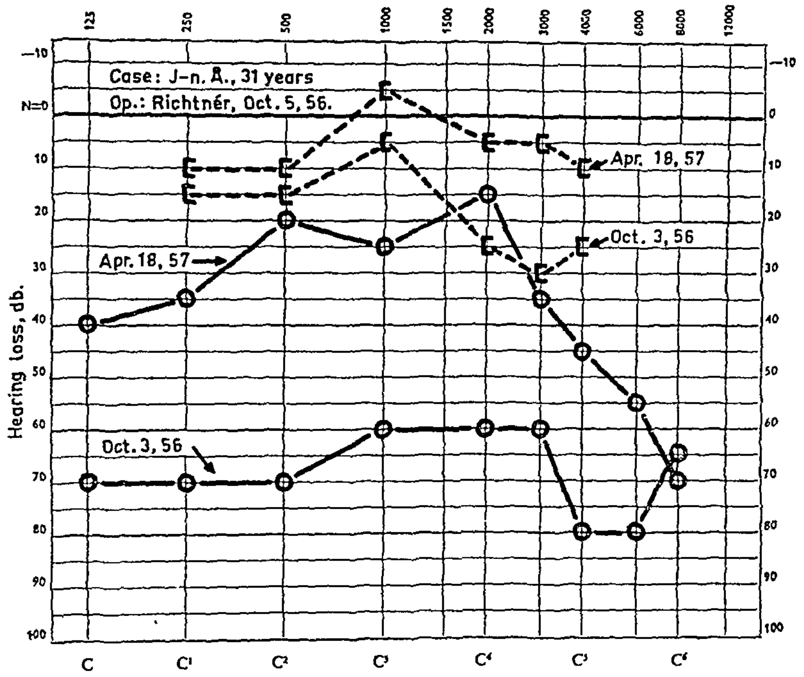

Fig. 3.

the patient's auditory acuity is 20 db as opposed to the pre-operative level of 63 db, in other words, an increase of 43 db. This strikingly good result is also difficult to explain audiologically inasmuch as the patient has only the stapes left. It is empirically useful to know that there are possibilities for achieving such good hearing after both malleus and incus are gone. This is good to know especially in cases of large cholesteatomas, where a conservative approach is not sufficient and where one is frequently forced to remove both malleus and incus. I will return to this problem later in this paper (see Fig. 3).

It is remarkably interesting that it is possible in cases of cholesteatoma to remove the cholesteatoma from the middle ear together with carious auditory ossicles and, by means of plastic screening of the round window, still retain hearing that does not differ greatly from the pre operative acuity, or, in some cases, even achieve an improvement

The following case is a beautiful illustration of the point I wish to make The patient was a 7 year old girl who had a long history of chronic inflammation of the right ear with a marginal perforation combined with osteitis and suspected cholesteatoma Her hearing was good and as mild an operation as possible was planned in order not to damage her hearing It was found, however, that she had a cholesteatoma that extended far into the middle ear about the ossicles and even down to and over the fenestra ovalis The incus was carious and the head of the stapes lay loose in the cholesteatoma Nothing of the posterior limb of the stapes remained The cholesteatoma extended even under the malleus and incus Consequently, in order to remove the cholesteatoma completely it was necessary to remove both the malleus and incus and it was decided to create a cavum minor The base of the stapes and the fenestra rotunda region were normal The tympanic ring was retained to provide a seat for the transplant Optimal closure was obtained with the flap over the hypotympanum No oxycel was used in the middle ear

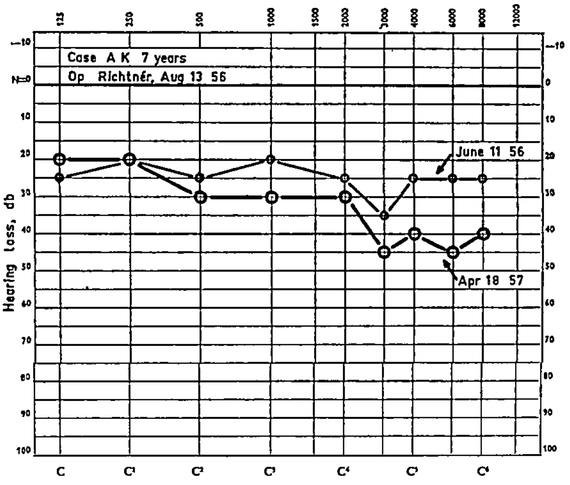

FIG 4

The patient's good hearing before the operation must have been due to a contact established by the cholesteatoma between the tympanum and the fenestra ovalis. In this case we were intensely interested to see what the auditory result would be. The middle ear healed well, and now, 9 months after the operation, there has been no occurrence of the cholesteatoma. Strange to say, this patient's hearing did not deteriorate greatly. Four months after the operation her auditory acuity was as good as 25 db and now, after 9 months, it is still at about the same level. Thus, with our operation, we have eliminated the cholesteatoma with removal of the ossicles and have still been able to conserve her hearing, thanks to this closure of the hypotympanum. The tubal passage is excellent. The auditory curve is shown in Fig. 4.

The material is too limited to permit any general conclusions on the effect of the treatment of adhesive otitis according to the cavum minor technique. Nevertheless, the results obtained would seem to indicate that we should continue along these lines. It is probable that we shall be able to form a definite opinion only after application of the technique for several more years. A broad field awaits the otologist.

We have now heard about the auditory results in the different individual groups. Taking the material as a whole we obtain the figures shown in Table I. The material comprises altogether 39 cases operated according to cavum minor technique, and almost half the cases have now been followed for one and a half years after the operation. The results of audiograms taken during the course of healing have been grouped as follows:

Group I comprises 37 cases with audiograms from 1 to 4 months after operation.

Group II comprises cases with audiograms from 5 to 9 months after operation.

Group III comprises cases with audiograms from 1 to 1½ years after operation.

We may say that throughout the patients subjected to operation had extremely pronounced auditory impairment.

As the Table shows, 37 patients were examined during the first four

TABLE I.

AUDITORY RESULTS IN ALL CASES AFTER TYMPANOPLASTY (CAVUM MINOR), 39 CASES

	1-4 months post-op.			5-9 months post-op.			12-18 months post-op.		
	Nb.	%	M.db	Nb.	%	M.db	Nb.	%	M.db
Improved	18	49	22	16	48	25	9	53	25
Status quo	18	49	—	17	52	—	7	41	—
Impaired	1	2	13	0	0	—	1	6	19
Total	37	100	—	33	100	—	17	100	—

M.db=mean decibels.

post-operative months. An improvement of more than 10 db occurred in 18 cases or 49 per cent. of the material. The mean improvement was 22 db. In 18 cases the hearing remained at status quo, i.e. there was neither improvement nor deterioration of more than 10 db. This represents 49 per cent. In one case there was a deterioration, and in that case it was 13 db.

During the period 5 to 9 months after the operation, 33 patients were followed, with 16 patients or 48 per cent. showing a mean improvement of 25 db. In 17 cases the hearing remained at *status quo*. No cases showed deterioration of hearing.

During the period from 1 year to 1½ years after the operation, 17 cases were followed. Nine patients showed auditory improvement. This represents 53 per cent. of the cases followed. The mean improvement was 25 db. *Status quo* may be reported in 7 cases or 41 per cent. One case showed deterioration. Examining these figures more closely we see that the mean improvement in hearing was already a mean of 22 db during the first four months after the operation and that the mean improvement after five to nine months and 12 to 18 months after the operation was 25 db. Thus, on the whole, the improvement measured in decibels was the same. This shows in the whole material that the improvement once attained with the cavum minor technique is not only maintained but even occasionally increased after five to nine and 12 to 18 months. On the whole, we may say the radically operated ears and the free transplants have healed well, and in almost 100 per cent. of the cases the tube is patent.

Prednisolone trimethylacetate (Ultracortenol, Ciba) has been a great help in the postoperative local treatment of the radical cavities. Tampons have been saturated with ultracortenol and packed in the radical cavities during operations and post-operatively. The inhibitive effect of prednisolone trimethylacetate on granulation formation is excellent. It has, moreover, proved an effective medium against infection.

I have nothing to contribute from my own material on the fifth form of tympanoplasty. I refer here to the cases where the aforementioned methods have failed, in which the stapes is fixed, and where a new fenestra is created in the wall of the horizontal semicircular canal. Perhaps, instead, in these cases an attempt should be made to mobilize the stapes.

CLINICAL RECORDS

LEUKOPLAKIA OF THE LARYNX COMPLICATED BY CARCINOMA

By J. SIEGLER (Liverpool)

CARCINOMA has for a long time been recognized as a complication of leukoplakia of the larynx. The case about to be described showed the clinical appearance of a carcinoma when first seen, but repeated biopsies, carefully examined, revealed only hyperkeratosis. However, after several years of careful observation, malignant disease was finally discovered.

Case History

G.G., aged 60, was first seen in *May, 1954,* suffering from intermittent hoarseness for years. On examination the larynx showed extensive ulceration of the right vocal cord almost involving the anterior end of the left cord. W.R. and chest X-rays were both negative. Direct laryngoscopy showed the right vocal cord to be thickened but biopsy showed hypertrophy of the squamous epithelium with chronic inflammatory changes of the sub-mucosa. There was no evidence of malignancy.

In *June, 1954,* direct laryngoscopy showed a growth extending over the whole right vocal cord, and biopsy showed the same changes as before.

In *November, 1954,* the right cord was still ulcerated but laryngoscopy and biopsy showed the same findings as before.

In *June, 1955,* there was a bulky hyperkeratotic mass on the right vocal cord and it was decided to treat him as though the lesion was malignant. A radical course of deep X-ray therapy to the larynx was commenced in *July, 1953,* the total dosage to the tumour was 6,000 r. over 42 days using the 250 kv. apparatus.

In *September, 1955,* the mass on the right vocal cord had disappeared completely and in *October* the right cord was reported to appear normal and to be moving normally. He was seen at monthly intervals but the inflammatory reaction resulting from the radiotherapy caused œdema of the mucous membrane over the arytenoid cartilages prevented adequate examination of the vocal cords, but in *October, 1956,* the œdema had subsided and the vocal cords appeared normal.

In *April, 1957,* he was hoarse again and the right vocal cord showed areas of leukoplakia involving the whole cord. These areas were partly papilliferous and biopsy showed squamous celled carcinoma. In view of the history and previous course of radiotherapy a total laryngectomy was performed.

Discussion

Leukoplakia may occur as a local or diffuse lesion of the larynx, usually on a vocal cord but occasionally elsewhere. The typical histological features are i.e. the keratinization with overgrowth of the surface epithelium with intact basement membrane and chronic inflammatory changes in the subepithelial layers in either variety.

It is significant that malignant change occurs so frequently although no figures are available to indicate the incidence but it has recently been suggested by Gordon (1950) that these patients are suffering from epithelioma *in situ*, i.e. a carcinoma within the layers of the superficial epithelium without any invasion of the surface or the basement membrane.

Larynx showing diffuse carcinoma of both vocal cords.

The problem of making an early diagnosis rests partly with the laryngologist in providing the pathologist with a sufficiently large biopsy which includes the subepithelial layers, and partly with the pathologist who must search through each section carefully for alteration in the shape of the cells and of their distribution as well as the shape and size of their nuclei. As identical histological changes occur in the superficial epithelium where there is an epithelioma *in situ* (Black, 1954).

It may be that the satisfactory response to radiotherapy which occurred in this case and has been reported in several cases in the past is due to the effect of the treatment on the carcinoma.

Summary

1. A case with carcinoma complicating localized leukoplakia is described.
2. The importance of detailed and repeated follow-up emphasized.
3. A suggestion has been made for the response in some cases of leukoplakia to radiotherapy.

I wish to thank Mr. J. McGibbon for his assistance in writing this case report and to Dr. Rewell and Dr. Harris for their pathological reports.

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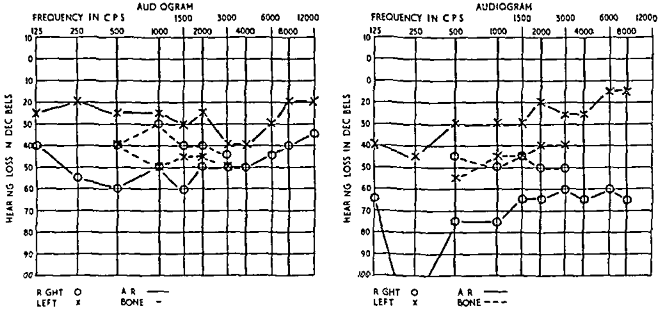
MALINGERING AS A FINE ART

By M E N SMITH (Swansea)

THE following case is presented for interest and amusement, though in the hope that the deeper implications, even if uncommented upon here, will provoke serious thought

A woman, age 22, was referred to the E N T Dept of Swansea Hospital by her doctor, on *January 23rd, 1957*, complaining of partial deafness and of pain in the right ear, which was alleged to have followed an accident at the brewery which employed her In this accident she claimed to have sustained an injury to the head part of some machinery having caught hold of her head in a vice like action, taking purchase by the vertex and below the mandible She was unconscious for 5 minutes



FIGS 1 & 2

January 23rd 1957 February 1st 1957

The patient was examined by a consultant E N T surgeon who found no clinical abnormality whatsoever in the ears, nose or throat, apart from a suggestion of reduced bone conduction in the right ear

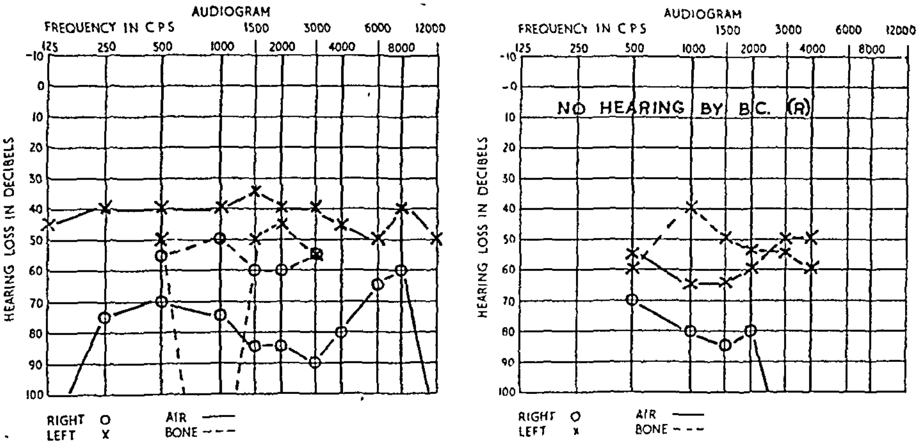
Audiometry was undertaken with the result shown in Fig 1

She appeared again on *February 1st*, stating that when she had looked in the mirror, upon rising the previous morning, she had noticed that she had two black eyes the right one being worse than the left Some bleeding from the right ear was noticed about the same time

Clinical examination appeared to confirm these statements, although it was noticed that the right tympanic membrane was intact and normal, and that the bleeding seemed to be coming from a laceration of the anterior meatal wall.

X-ray of the skull and sinuses, taken the same day, showed no evidence of bony injury.

The patient's next attendance was on *February 13th.* The appearance of the eyes was unchanged and, in view of the lapse of time, this naturally aroused the gravest suspicions in the mind of the examining surgeon. These suspicions were confirmed when it was found that the "ecchymoses" could be completely removed with a wet swab! It appeared that some substance such as ink, or perhaps "eye shadow" had been applied with consummate artistry to produce the desired effect. The patient, however, was quite unmoved and nonchalantly informed us that the bluish substance was an ointment supplied by her general practitioner.



FIGS 3 & 4

February 13th, 1957 February 22nd, 1957

In the light of these developments, a series of audiograms was prepared in an attempt to test the validity of the alleged deafness. These are shown in Figs. 2, 3, and 4. Considering the four audiograms together, it will be seen that they are all different, although the first three do follow roughly the same pattern. The actual hearing levels shown, are, however, scarcely compatible with the fact that the patient hears normal conversational voices at ordinary social distances without the slightest difficulty. Similarly, it is scarcely possible to accept her claim that she is quite unable to hear a 512 c.p.s. tuning fork, maximally loud, placed on the vertex, or anywhere else in the sagittal plane of the skull—a claim which incidentally renders the Chimani-Moos test inapplicable.

The fourth and final audiogram was carried out when the patient was seen for the last time on *February 22nd.* This bore little or no resemblance to its three predecessors At this visit there was a further issue of blood from the right ear and otoscopy suggested that the original laceration had been re-opened to produce it

Upon the evidence here presented the patient was regarded as a case of simulated deafness, the aural hæmorrhage was taken to be due to a self-inflicted injury, while the "black eyes" were a final cosmetic touch for full measure At this stage, and without indicating our conclusions to her, we referred the patient back to her general practitioner, who was, of course, provided with a full report So far, nothing further has been heard of the matter It will be observed that if this patient had confined her complaint to the original one of deafness and desisted from the later embellishments, she might well have succeeded in her object, especially in the hurly-burly of a busy out patient department

It only remains to say, that at a late stage of our investigations the patient herself broached the possibility of a claim for compensation against her employers, on the basis of her injuries sustained while at work This, we thought confirmed our conclusions, had any confirmation been needed

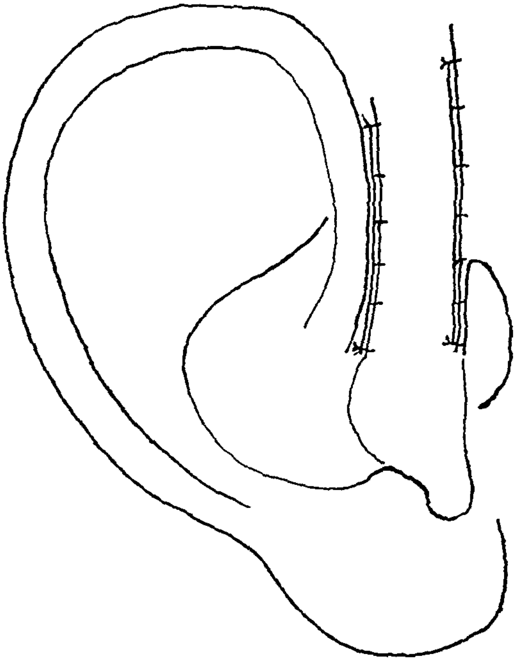
I wish to thank Mr J Crowther, whose patient this was, for permission to publish, Dr Mandl, Consultant Radiologist, for X-ray interpretation and discussion and for implanting the germ of suspicion in my mind, and Sister Watts of the E N T Department, for audiometric assistance and support in being, like myself, completely taken in by the "black eyes" when we first saw them

CLINICAL NOTE

THE SUPERIOR VIDAU FLAP

By R. L. FLETT (Derby)

THE Vidau (1954) pedicle flap has been a great help in securing the blood supply to the skin graft in a modified radical mastoid. *Fortunato* (1954) has been accustomed to take a large flap of skin from the posterior surface of the pinna over the mastoid, and then to perform a tympanoplasty through the post-aural incision. Owing to convenience, the endaural incision has been persisted with in my department, even although skin is removed from the back of the auricle.



The Vidau flap has been used on a number of occasions and the only disadvantage seems to me, that there is still a raw area from the introduction of the flap from the posterior wound. It has, therefore, been modified and the flap attached superiorly instead of inferiorly, and the first half-inch of the pedicle is made into the form of a tube.

The supra-meatal part of the incision is sewn up and to avoid tension on the flap, in the region of the middle ear, the skin flap can be sewn at the beginning of the bony meatus to the skin of the external auditory meatus

Instead of the tube pedicle, the base of the graft can be stitched into the upward continuation of the endaural incision (diagram) This does away with the necessity of forming a tube, though the tube undoubtedly does make sure of an adequate blood supply to the flap, in that a small artery forms inside a fortnight There is, however, a branch of the superficial temporal artery which runs down the posterior surface of the pinna, and if this can be left intact, and not divided at the junction of the pinna and the scalp, then there is a necessary blood supply to the flap The upper inch and a half of the flap need not be thinned down very much until near the level of the attic region The cavity is packed with gel sponge and as it is totally lined by skin, can be left for a fortnight Then the patient is given another anæsthetic, the sutures removed and the gel sponge can be sucked out carefully so as not to detach the lower end of the flap Usually this leaves a cavity which is totally lined by skin, and which requires no postoperative attention

The flap lies better over the drum region if the apex, very thinned out, lies in the floor of the meatus The sutures are necessarily very small, but with the use of antibiotics continuous atraumatic nylon sutures can be employed Another advantage is that a large area of the skin can be removed from the lower part of the mastoid and pinna and there is not often need to take a further Thiersch graft

Retraction and scarring cause the tragus and helix to approximate and not leave the large gap as shown in the diagram

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SOCIETIES' PROCEEDINGS

ROYAL SOCIETY OF MEDICINE—SECTIONS OF LARYNGOLOGY AND OTOLOGY

COMBINED SUMMER MEETING AT THE
UNIVERSITY OF BRISTOL

President—PHILIP J. JORY, D.S.O., F.R.C.S.

Thursday, Friday, Saturday, June 27th, 28th and 29th, 1957

Plastic Surgery of the Middle Ear

THE opening paper by Mr. Mawson is to be found on p. 56.

Discussion

MR. F. MCGUCKIN: The opening speaker has given us much to think about and all are to be congratulated on their presentation, I would take up one or two points for comment.

Time-consuming investigations on the hearing may be of some practical value in the absence of active disease, but they can be misleading where disease is active. Most of us have seen cases in which a small ball of cholesteatoma has acted as an efficient incus and indeed Dr. Richtnér has mentioned such a case. The working clinician with little time to spare must decide which of these investigations are valuable and when they are necessary.

There has been almost no reference to the condition of "the other ear" and in my view if there is one sound ear, this fact will often determine what advice we will give to the patient. If there is one good ear I do not assess any functional improvement which is short of approximately the 30 db mark as a clinical success though it may be a technical achievement.

At the March, 1955, meeting of the North of England Society held in Newcastle about two dozen chronic otitic cases were shown. These were presented in four groups:

Group 1

Cases in which attempted reconstruction of function had been wholly successful—for example, an attempt at complete reconstruction had produced normal leverage plus normal sound protection of the round window.

Group 2

Cases in which the functional result was only partly successful—for example, an attempt at columellar effect turned out to be no better than a simple baffle over the round window (Wullstein's Type 4) We now have various titles for this Type 4 result, including cavum minor, kleine pauche, petite caisse, and round window baffle

Group 3

Cases in which the functional result could only be classed as a failure, with no useful leverage and an inefficient or absent sound protection

Group 4

Cases in which no useful reconstruction was considered possible—an extreme example would be that of invasion of the labyrinth by cholesteatoma

I still think that this is the most critical way of looking at these newer techniques, and in order to be critical one must relate results to the material which comes our way It is worthwhile noting that of 70 major cases which I have had to undertake in 17 months nearly a quarter were already suffering from invasion of the labyrinth, and about one in three showed exposure of the facial nerve Nevertheless, in this material I attempted some kind of primary reconstruction in about half I am not disappointed that the functional failure rate has been high because the material has been of just about the worst possible kind There are reasons why my sample of cases is much worse than the average, reasons why I should have had in this 17 months only two cases which were inactive at the time of operation, but these I hope to present at a future meeting

Mr Gavin Livingstone stated that the reconstructive surgery of the middle ear should not be undertaken until all pathology has been removed at operation, and only after this should the problem of plastic surgery reconstruction of the middle-ear cavity be considered

In his opinion the word "myringoplasty" should be discarded and the operation called Tympanoplasty Type I, as this operation is identical to the myringoplasty, and in all the types of reconstructive surgery some form of skin graft to the drum has to be undertaken, and Zollner s classification covers all types

In the cases where there is a large central perforation and the ossicular chain is broken, simple patching of the drum, as advocated by Mr Mawson, is, in his opinion, the wrong operation, and a Tympanoplasty Type III should be attempted and a columellar effect obtained Out of 113 mastoid operations done during 1955 and 1956, the Tympanoplasty Type I was done in only 20 cases, while Types II and III were done in 65 Twenty eight cases had atticoantrotomies in the earlier months, but I found no improvement in hearing, and he does not do this operation now, as some type of plastic reconstruction is applicable in every case of chronic otitis media where operation is indicated

Mr I B Thorburn (Blackpool) thought that it was necessary to review some of our established surgical principles underlying the treatment of chronic otitis media A careful microsurgical technique now made it possible to assess pathological changes in the tympanum as irreversible or reversible Reversible pathology referred to reactive mucosal swelling and recognized the possibility that when conserved it might resolve to form a healthy mucosal lining to the new tympanic cavity. Irreversible pathology referred to cholesteatoma, keratinizing squamous epithelium, granulations or osteitis and must be eliminated. The use of meatal skin flaps to line part of the tympanic cavity as had been described by Mr. Mawson seemed contrary to this principle. Similarly, he disliked the idea of using Thiersch grafts in a closed fenestration operation as he believed was advocated and practised by Mr. Simson Hall.

Mr. Thorburn thought that Wullstein's apparent insistence on retaining a bridge as a condition of carrying out a type II tympanoplasty was regrettable and possibly misleading. It seemed to follow that if it were necessary to remove the bridge in order to deal adequately with the underlying disease then one should also remove the incus and head of malleus and complete a type III tympanoplasty. Especially in cases of attic cholesteatoma with good initial hearing by removing the bridge one could gain access to disease yet retain an intact functioning ossicle chain and reconstruct a closed tympanic cavity with or without the help of a full thickness skin graft. In this sense a type II tympanoplasty without a bridge was similar to a grade 2 modified radical mastoid operation, and in his experience these cases usually retained very satisfactory hearing.

DR. SIMSON HALL said: Results of operation are the indications of its success and Mr. Mawson's results in myringoplasty can hardly be improved. There is no doubt that selection of cases plays an important part in this success and now I always employ the method of inspection of the aditus and antrum by a window as is recommended by Wullstein. This enables an accurate assessment of the situation to be made and the operation planned accordingly. The method described of turning in a flap from the meatus I consider useful and contrary to expectation does not result in cholesteatoma formation. Epithelium can be used in cavities without fear.

MR. COLLINS detailed a case of attico-antrotomy operated upon by the late Mr. John Gerrie. This patient developed a cyst in the aditus which occasionally discharged and as she was complaining of a certain amount of giddiness, operation was indicated. In actual fact, the cyst contained only a little glairy fluid but it was thought advisable to explore the middle ear by turning back the tympanic membrane. There was no cholesteastosis in the middle ear but direct visual proof was obtained that the reason why this patient had such good hearing was that these were adhesions between the tympanic membrane and head of the stapes. This patient showed only a 20-30 db loss in hearing following her attico-antrotomy.

J. F. O. MITCHELL (Dundee) said: I should also like to compliment Mr. Mawson on his results, but should like to ask how long he requires his patients in hospital, and whether this is really necessary.

We must not forget what work has already been done with caustics and how this can be modified today, for perforations not marginal or actively infected.

I have recently completed a series of 50 cases using trichloracetic acid cautery followed by the application of a patch of gelatin sponge soaked in autogenous blood-clot. Thirty-six of these cases have healed well, and 14 of these by a single application as an out-patient, with little time or trouble on the part of surgeon or patient; at the same time the patch gives an immediate assessment of the hearing improvement, and it is surprising how large a perforation will heal by this conservative method.

In many cases hearing-loss is not the problem, but social freedom to swim and shampoo, and fitness for military service; and surely conservatism can first be tried as an out-patient, reserving myringoplasty for the failures after two or three attempts.

E. H. RAINER showed the audiogram of a girl of 14 who had an attico-antrotomy done in 1951 for extensive cholesteatomatous disease Her average loss is only 10-15 db.

This illustrated how good the hearing can on occasion turn out to be, even before the benefits of tympanoplasty were known.

GENERAL NOTES

THE Editors are anxious to improve the clinical aspect of the Journal and would welcome more reports of interesting cases. These clinical records need not be at all elaborate and it would be unnecessary to go minutely into the ætiology or the literature of the subject. Senior Registrars have unique opportunities for seeing a large variety of cases, many with unusual features. It is in many ways regrettable that these should go unrecorded. It is hoped that all those in charge of clinics will help us in this matter and see that such cases are not overlooked.

BRITISH POSTGRADUATE MEDICAL FEDERATION

COURSE OF LECTURES ON THE SCIENTIFIC BASIS OF MEDICINE

THE following lecture will be delivered in the Lecture Theatre of the London School of Hygiene and Tropical Medicine on Thursday, February 6th at 5.30 p.m.:—

Dr. C. S. Hallpike—"Some remarks on the scientific basis of otological practice."

OTOLARYNGOLOGY LECTURES 1957-58

THE following lectures, arranged jointly by the Royal College of Surgeons of England and the Institute of Laryngology and Otology, will be delivered in the Lecture Hall of the College, at 5.30 p.m.

1958
February 6th. Mr. R. H. Franklin. Congenital abnormalities of the œsophagus.
March 6th. Mr. H. Jackson Burrows. Disorders of the cervical spine.

May 1st Professor W. D. M. Paton The pharmacology of mucous membrane
July 3rd. Mr Norman C Tanner Haemorrhage from the Œsophagus

INTERNATIONAL CONGRESS ON THE MODERN EDUCATIONAL TREATMENT OF DEAFNESS

An International Congress on the Modern Educational Treatment of Deafness will be held at the University of Manchester from July 15th-23rd, 1958

The Congress is being organized with the approval and support of the Council of the University, with financial help from the Ministry of Education, the National College of Teachers of the Deaf, and the Rathbone Trustees, and with the co operation of the British Council and the Alexander Graham Bell Association for the Deaf, Inc , Washington, D C , U S A

The Congress is being planned to bring together educators, otolaryngologists, pædiatricians, medical officers, audiologists and educational psychologists, to consider and discuss the significance, to the future development of their work, of the results of modern research on problems of deafness in childhood

Arrangements are being made to offer accommodation to members of the Congress in University Halls of Residence The cost of this type of accommodation is expected to be about £1 2s per day, including bed, breakfast and dinner The University Refectory will be available for meals during each day

Forms of application for membership of the Congress will be supplied upon request, to Professor A W G Ewing, Department of Education of the Deaf, the University, Manchester 13, England

INSTITUTE OF LARYNGOLOGY AND OTOLOGY

330 Gray's Inn Road, London, W C 1

The Academic Board announces the following short course which is intended for Consultants and Senior Registrars —

"Modern methods of testing the auditory and vestibular systems"

Wednesday, February 12th The Auditory System

9 30-1 0 p m Pure tone and speech audiometry, recruitment, limen difference, Bekésy technique, probe testing
2 0 4 30 p m Practical demonstration of methods

Thursday, February 13th. The Vestibular System.

9.30-1.0 p.m. Normal response to positional, optokinetic, rotary and caloric stimulation.
Nystagmography—electrical recording.

2.0-4.30 p.m. Practical demonstration of methods.

Applications for enrolment should be sent to the Dean. Fee—£3 3s. (including morning coffee, lunch and tea).

The Journal of
Laryngology and Otology

(*Founded in* 1887 *by* MORELL MACKENZIE *and* NORRIS WOLFENDEN)

February 1958

THE MOBILIZATION OPERATION IN THE TREATMENT OF OTOSCLEROSIS*

By I SIMSON HALL (Edinburgh)

Introduction

STUDY of the papers written by James Yearsley show that his chief interests centred round the relief of middle-ear deafness, especially of the catarrhal type as opposed to the suppurative It seems that a great many of his patients must have been otosclerotics who even at the present day are still too often given this "catarrhal" label It therefore seemed appropriate that this lecture should be concerned with modern treatment directed to the same ends and should thus supplement and continue the work of the founder of the Metropolitan Ear, Nose and Throat Hospital

Historical

Since Rosen rediscovered the possibility of freeing the stapes by direct attack, interest in this procedure has been intense and world-wide Why this once popular procedure should have been completely forgotten is a fascinating speculation which is rendered all the more interesting when one finds that the operation was extensively practised by a number of surgeons Miot's classical study was published in 1890 and at that time others, namely Schwartze, Boucheron and Gelle, had their own methods, but they do not appear to have published on the subject

Miot reported 125 cases, claiming improvement in 74, but it is worth noting that the operation was performed for other conditions besides otosclerosis. It is difficult to be certain which cases were otosclerosis and which the result of suppuration.

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* Based upon the James Yearsley Memorial Lecture delivered on 28th February 1957 in St Mary Abbots Hospital London

It is interesting to find, however, how complete and careful his operation seems to have been. Cocaine produced the local anæsthesia and considerable care was taken with the antisepsis of the external meatus.

The exact method of approach probably changed but the recommended route was an incision along the posterior edge of the drum, and was called a "myrindectomy".

After such a thorough trial and after results which appeared to claim 60 per cent. of success, it is at first sight difficult to understand why this operation was discarded so completely, and indeed forgotten.

There is no doubt that dissatisfaction caused other workers to continue searching for the answer to the otosclerotic problem in other directions, and they gradually evolved the modern fenestration operation.

There is probably a parallel to the development of the fenestration operation. Jenkins, Blake, Bárány, Fraser and others abandoned the fenestration operation owing to the pressure of public opinion which held that the operation was too dangerous and therefore unjustifiable. This seems the most rational explanation in the case of the mobilization operation too, for a 60 per cent. improvement rate was not good enough to justify even a few cases of acute middle-ear disease, amongst which may possibly have been one or two fatal cases of meningitis.

The introduction of antibiotics is the factor which today makes the difference between our success and the failures of our predecessors.

Obvious advantages of this technique are, the shortness of stay in hospital, the absence of anæsthetic risks, and the small loss of working time. Points such as these make further development and experiment well worth while, in spite of the comparatively low rate of success.

Already there are many variations in the treatment of the stapes and it is as well to consider what it is we are attempting to do.

Helmholtz was the first to demonstrate the movement of the ossicles, and he showed clearly that the movement of the stapes was so small as to be almost invisible without a microscope.

In this operation the stapes is moved about in a manner in which it was never intended by nature that it should move. The treatment is crude in the extreme, but occasionally successful. By this rough usage an attempt is made to disrupt the otosclerotic process which is fixing the stapes.

Pathology

There are two factors governing the fixation of the stapes. First is the stage at which the otosclerotic process happens to be in its development, and second, the amount of involvement of the stapes itself. Whether the process is a localized one or whether it is diffuse, is also important.

It has been shown elsewhere that otosclerosis is a healing process in which nature is making good an early defect in the bone From these studies it is obvious that healing and quiescence of the disease can take place at any time and that only a small area of the capsule may be involved Again, it may be almost a generalized condition with exostosis and marked thickening and overgrowth of the bone

If healing is slow or just commencing it is unlikely that any breaking down of the newly formed trabeculae of bone will have a permanent effect upon the symptoms of the disease

On the other hand if the process is advanced and the bone fusion between the stapes and the window is complete, stable and well-formed, it is easy to understand how it is possible to fracture sharply this union and then to expect the fracture to persist as a non-union between the bone edges

Many workers have demonstrated that fracture of the bone of the labyrinth capsule does not regenerate as bone but unites by fibrous tissue, and it is reasonable to believe that this is the result in such cases as are dramatically successful

This conception of the pathology of the disease provides the only reasonable explanation of the success or failure of the manœuvre

Methods

The problem therefore is to break through the bony bridge which unites the stapes to the margin of the oval window, or to fracture the footplate so that a false joint is created behind the fixation point The method of doing this is obviously not of any importance provided that irreparable damage is not done to the ossicular mechanism Whether we use pneumatic hammers, rotatory vibrating burrs, or a simple mobilizer as recommended by Samuel Rosen, is immaterial

A study of the wide varieties of the types of fixation, such as that carried out by Professor Ruedi on the Zurich Collection, has shown how widely the manner of fixation can vary It would seem that the differences in the pathological process are so unpredictable that it will have to be accepted that a proportion of cases will be failures

Samuel Rosen has suggested a way to overcome some of these failures, by boring a small hole in the edge of the footplate which is sufficient to remobilize the labyrinthine fluid

Experience of this technique is limited and therefore any results which are quoted must be regarded as purely preliminary

Results

In all 172 operations have been carried out, and broadly speaking the fenestration operation is not performed until the result of mobilization is known. Exceptions to this general rule are such cases as those who come from a considerable distance expressly for a fenestration and to whom time is material and who cannot afford to return in case of failure. Some patients too are attracted by the higher success rate and prefer the greater certainty of the fenestration operation.

The chief objection to carrying out the fenestration at a very early date is that improvement in hearing may not be achieved for some weeks after mobilization. It is wiser, therefore, to wait for possible improvement.

TABLE I.

TOTAL NUMBER OF OPERATIONS 172.
IMPROVEMENT OF 10+ DECIBELS.

Class 1	50 per cent.
Class 2	65 per cent.
Class 3	28·5 per cent.

TABLE II.

TOTAL NUMBER OF OPERATIONS 172.
IMPROVEMENT OF 15+ DECIBELS.

Class 1	46 per cent.
Class 2	51 per cent.
Class 3	6 per cent.

Tables I and II show the results obtained in 172 operations classified into the usual groups. It is noteworthy that the greatest improvement is shown in Class 2 and not in Class 1. This is in keeping with the conception of the pathology of the disease already stated, for Class 2 is composed of older and more established cases in which the bone is more likely to be healed, while the more immature types of otosclerosis are to be found in the younger age groups and the earlier forms of the disease.

The case of very long standing with some evidence of nerve deafness is a very poor prospect, possibly because in this case the involvement of the stapes is great and the length of time and the degree of consolidation of the bone has rendered fracture of the surrounding bone impossible. The greatest improvement which I have obtained is 40 decibels, and in no case has the hearing returned to normal. Why this should be is difficult to understand because the ossicular chain is intact and the stapes has presumably returned to full mobility. The explanation is probably that it is almost impossible to produce by mobilization the free movement of the unrestricted stapes and so some residual loss of movement must remain. Whether this will prove eventually a satisfactory explanation remains to be seen.

Movement of the Stapes

It is essential that there should be a clear idea what should be done to the stapes in order to free it and break down pathological bone.

The stapes is attached to the oval window in such a way that the upper edge moves to a greater extent than the lower and the anterior edge more than the posterior. The movement of the upper edge, which is the chief movement, is largely inwards though at the same time the lower edge would move fractionally outwards. Any movement, therefore, applied to the stapes should as far as possible simulate the normal in at least the first instance. As the head of the stapes moves inwards it also moves upwards to some extent. Pressure over the head of the stapes directed inwards and slightly upwards should therefore be the first attempt. This has the advantage in applying force in the direction in which the crura can best support it. This pressure can be applied either directly to the stapes or through the lenticular process of the incus.

If these first attempts do not meet with success then greater pressures must be exerted in other ways, always keeping in mind which direction is the normal.

The footplate is so fixed at the lower edge and posteriorly that there is little point in trying to move it in these parts against the normal anatomical attachments. This point is so fundamental to the whole operation that it is worth remembering throughout this stage.

If these normal movements fail then one of the other techniques must be considered.

Helmholtz has made some interesting observations on this point and shows that even in face of extreme movement of the malleus the incudo-stapedial joint is not disturbed; in fact the normal place for the lenticular process to be is in contact with the head of the stapes. Therefore even if one disrupts the incudo-stapedial joint it is probably sufficient to place the bones in contact to obtain reunion.

It is a matter of practical experience that a good result can be obtained when it is known that the joint has been injured.

Technical Details

Local anæsthesia by means of 1 per cent. Xylocaine with Epinephrine delivered from a dental cartridge syringe is used. As a rule about three-quarters of one cartridge, 1 c.c., is sufficient to give complete anæsthesia.

Three points are injected: roof, posterior meatal wall, and floor. In each case injection is made down to bone at the outer rim of the external bony meatus. No attempt is made to infiltrate the canal wall as this may cause the fluid to run under the epithelium and balloon the surface epithelium of the drum. This makes it exceedingly difficult to place the incision accurately and to make it without tearing off the outer layer from the drum.

The microscope is used during the whole operation, as the available field is the same for glasses and microscope, the only difference is the degree of magnification. The anterior bony wall of the meatus limits the field for both instruments alike when making the incision and thereafter the limitation is regulated by the handle of the malleus and the reflected drum.

The incision used for this operation is the "eleven to six" incision in the right ear, with the incision furthest from the drum at about "nine" and returning to the drum membrane above and below.

Should the incision be made close to the drum membrane all the way, and it is found necessary to remove part of the bony edge of the tympanic ring, difficulty may be experienced in covering the defect unless an adequate amount of meatal skin is available.

There is no doubt that individual preference plays a great part in deciding what instruments will be used for different manœuvres and so it is not suggested that any particular instrument is more suitable than another.

A rounded sharp instrument is the most suitable for making the initial incision. Whether it is one of those designed by Dr. Rosen for bone removal or some other, a rounded instrument fits better the concave surface of the external canal and is not so likely to tear the skin as is a sharp pointed knife. This applies also to elevators, and such as are square at the corners cannot be brought to bear over any length of their edge. It is also essential to resharpen all instruments after each operation.

Elevation of the flap may be easy or difficult. It is safer to commence in the floor as this is the thinnest and most delicate part and most liable to tear if pulled upon before fully detached.

When the edge of the tympanic ring is approached the drum can be detached from its sulcus, and the bluish membrane of the middle-ear lining is seen and is the sign that elevation is complete.

At this point, to ensure that hæmostasis is complete, pledgets of wool soaked in Adrenalin are inserted and held in position until all bleeding ceases.

The middle ear is then opened, usually with the seeker or some such sharp instrument which separates the lining membrane from the drum membrane along the edge of the tympanic ring. The drum membrane is then folded forward and exposes the contents of the middle ear.

The chorda tympani comes readily into view and in many cases appears to interfere with access, but it can be pushed aside, and it is rarely necessary to divide it. With increasing experience it is frequently possible to do all that is required through a smaller opening. The stapes is then tested with the seeker for mobility as recommended by Samuel Rosen.

There should be no difficulty in identifying fixation as experience with the normal should have given the necessary "feel" to this manœuvre.

Inspection of the middle-ear cavity including the niche of the round window should be carried out as far as the access described will permit. Adhesions, exostoses, and the like are the conditions sought.

Testing Hearing

Hearing is tested by means of the audiometer and progress if any can be judged accurately only in this way

Apart from the usual pre operative studies the hearing is tested after the drum has been reflected and the stapes exposed, so that any improvement after moving the stapes is immediately apparent If the improvement is not satisfactory then mobilization is continued and the test repeated

Another method of judging mobility of the stapes is to watch a light reflex in the round window If this moves synchronously with the induced movements of the stapes then obviously there must be movement within the labyrinthine fluid and the object of the operation has been achieved This sign has to be used with the greatest care, for the very tiny head movements caused by the movement of hand and wrist are sufficient to cause apparent movement of reflexes when seen through a microscope at ten diameters Probably pulse and breath movements may have a similar effect

On many occasions although good movement is obtained there is no real improvement in hearing This may be due to the adhesions between the footplate and the oval window being widespread and deep and not wholly ossified

Occasionally there is evident thickening of the membrane covering the edge of the oval window and though the footplate can be seen moving throughout its length there is no real improvement in hearing

The most successful cases are those which respond most readily and quickly to mobilization, sometimes even with the small click which can be so readily confused with the sound of the fracturing crus

Where a satisfactory result appears to have been obtained the drum membrane is rolled back into place and the incision is covered by a small slip of oil-silk upon which is pressed a pledget of optrex tulle or other packing The hearing rises about 5-10 decibels on replacement of the drum membrane

After-treatment comprises the administration of Penicillin as cover The pledget is removed in one or two days and the oiled silk in four or five These times are not critical and it is found that the drum membrane returns to normal in about fourteen days

Complications

The diagnosis may have been wrong as with a case recently operated upon in which the drum was apparently normal except for a small scar in the posterior part In this case the middle ear was full of thick tenacious mucus, which was only removed with the greatest difficulty with powerful suction

The middle ear may be found full of adhesions covering and enveloping everything. These are of two kinds, those which are the result of previous middle-ear suppuration and those due to lack of absorption of embryonic tissue in the middle ear. In the first case it is unlikely that the condition was unsuspected as the history or appearance may have prepared the operator for what was to be found. The other type however may give no sign before the ear is opened, and then the operator has to clean the ossicles and the windows carefully of the many thin clear adhesions.

The instruments most useful for this are the small hooks used for clearing the canal of shreds of endosteum after the creation of the fenestra.

This condition is probably part and parcel of otosclerosis. Just as the otosclerotic process is a disturbance of mesenchyme, so this is likewise residual mesenchymal tissue and accompanies otosclerosis. This probably accounts for the fact that the so-called chronic adhesive process has been shown to be amenable to cure of the resulting deafness by fenestration.

Tearing and perforation of the drum membrane is always a possibility, but rarely persists. It is partly to guard against trauma or adhesion of any packing to a damaged drum that oil-silk is used to protect it after operation, for if perforation is suspected the oil-silk can be left in position for a prolonged period and the drum will almost invariably heal underneath the silk.

So far only one case of persistent perforation has been encountered, where a previously latent otitis externa flared up after operation in spite of all antibiotics and resulted in a small perforation.

Facial paralysis occurred on one occasion only, due to infiltration of the local anæsthetic, and it cleared away completely as the anæsthetic wore off.

Indications and contra-indications

Contra-indications are few, but include local conditions such as obvious scarring and damage to the middle-ear structure by previous suppuration. Catarrhal conditions and sinusitis should be attended to just as in the case of the fenestration operation.

As far as indications are concerned one may say that all patients suitable for fenestration are suitable for mobilization, but not all suitable for mobilization are suitable for fenestration.

In the first place one would undertake this small procedure in cases which would be considered as not sufficiently advanced for fenestration, and in unilateral cases in which a fenestration would not be justified. Also where only one ear offers any hope of improvement, as damage and complications are so rare, it is justifiable to carry out the mobilization operation.

At the other end of the scale some patients regarded as hopeless prospects for fenestration can be helped to make better use of their hearing aids.

In cases of doubtful etiology a direct attack on the stapes may clear up the diagnosis and ascertain suitability or otherwise for fenestration.

To sum up the situation it seems that there is here a real contribution to the treatment of otosclerosis, and one which is likely to be of increasing value as we learn more of its uses and limitations.

THE FENESTRATION OPERATION: A REVIEW BASED ON AN ANALYSIS OF 139 OPERATIONS

By R. B. LUMSDEN* (Edinburgh)

OF 205 fenestration operations performed on 200 patients, 176 were carried out from 2 to 10 years ago. Follow-up has been possible in 136 patients, 3 of whom had bilateral operations, giving *a total of 139 operations which form the basis of this communication.*

Sex. 93 were females and 43 males. These relative figures are of no significance as they were largely determined by availability of beds.

Ages were between 17 and 56, the average being 35. 72 cases were between 17 and 35 and 64 cases between 36 and 56.

The duration of deafness varied from one to 30 years. The presence or absence of a *family history* of deafness was recorded in 121 cases. A significant positive family history was present in 49 and absent in 72 cases.

The operative technique employed essentially followed that described by Simson Hall (1944, 1945, 1946) with minor modifications. The mortality rate was nil.

Classification and Assessment of Results

As in a previous communication (Lumsden, 1949), cases have been classified, according to the pre-operative findings, into three groups—favourable, borderline or doubtful and unfavourable (after Passe, 1947).

1. *Favourable.* Those in which clinical and audiometric examination show no appreciable degree of inner-ear deafness, pure tone audiometric examination by air conduction showing that hearing loss in the speech frequencies does not exceed 60 db.

2. *Borderline.* Cases in which deafness is of such degree that it is necessary to obtain the maximum improvement if serviceable or socially adequate hearing is to result. On pure tone audiometric examination the loss at one or more of the speech frequencies by air conduction may exceed 60 db., but the loss by bone conduction in the speech frequencies (representing impairment of inner-ear function), after making the necessary corrections (Shambaugh and Carhart, 1951), does not exceed 30 db.

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3 *Unfavourable* Cases in which the associated inner ear deafness is of such degree that the chances of success are practically nil The net loss in the speech frequencies by bone conduction exceeds 30 db

Results Cases are classed as *successful* when all three of the following requirements are fulfilled (i) the patient is satisfied that serviceable or socially adequate hearing has been obtained (ii) confirmation of this is obtained by testing the patient s ability to hear the conversational voice and the whisper (iii) *the average hearing loss by air conduction in the speech frequencies on the pure tone audiometer does not exceed 30 db at the end of a period of 2-10 years after operation*

Speech audiometry is not considered since it was not available when the present series of cases were operated on

It is realized that the foregoing criteria are open to criticism, but it is claimed that they afford a consistent analysis extending over the ten year period

Results

When the 139 operations are placed in their appropriate groups, results are as follows

HEARING RESULTS 2 10 YEARS AFTER OPERATION

Pre operation Group	Number of Operations	Successful	Improved	Unchanged	Worse
1 Favourable	92	73-79% (89%)	9=10%	6= 7% (11%)	4= 4%
2 Borderline	30	6=20% (77%)	17=57%	5=16% (23%)	2= 7%
3 Unfavourable	17	3=18% (41%)	4=23%	7=41% (59%)	3-18%
All groups	139	82=59% (81%)	30=22%	18=13% (19%)	9- 6%

FAVOURABLE GROUP ONLY HEARING RESULTS 3 6 MONTHS AFTER OPERATION

Number of Operations	Successful	Improved	Unchanged	Worse
92	80=87% (92%)	5= 5%	3- 4% (8%)	4= 4%

Within the favourable group, a comparison of results has been drawn in terms of *lower and higher age groups*

	17-35 years	36-56 years	Totals
Successful	38	35	73
Improved	5	4	9
Unchanged	4	2	6
Worse	4	0	4
	51 +	41 =	92

The only significant finding appears to be that all of four results in the "worse" category fall within the lower age group.

Tinnitus

The presence or absence of pre-operative tinnitus was recorded in 109 cases. It was present in 84 and absent in 25. A comparison of operation results suggests that tinnitus *per se* is of no particular significance in relation to hearing prognosis.

Post-operative tinnitus has been absent in the operated ear in the great majority of cases, irrespective of the hearing result. In a few, it has been only modified and in only one has it continued to be a cause for complaint, in spite of a successful hearing result. One patient successfully sought operation on the second ear with the main object of obtaining relief from tinnitus.

Paracusis

The presence or absence of paracusis was recorded in 106 cases. It was present in 80 and absent in 26. A comparison of operation results is shown below. This does not suggest that paracusis *per se* is of favourable significance:

HEARING RESULTS IN RELATION TO PARACUSIS.

	Pre-operation Group	Number of Operations	Successful	Improved	Unchanged	Worse
Paracusis +	1. Favourable ..	56	45=80% (89%)	5	4	2
	2. Borderline ..	15	3	8	3	1
	3. Unfavourable	9	1	3	4	1
	All groups	80	49	16	11	4
Paracusis −	1. Favourable	19	16=84% (95%)	2	0	1
	2. Borderline	3	1	2	0	0
	3. Unfavourable	4	1	0	2	1
	All groups	26	18	4	2	2

Pregnancy

Information regarding pregnancy has been recorded in 20 cases, 18 of which had at least one child *previous to operation.* 12 cases reported that deafness commenced, or became appreciably *worse*, during pregnancy.

The operation results are as follows:

Favourable group	5	Successful	4	(Onset after second of 3 pregnancies in one)
		Worse	1	
Borderline group	4	Improved	3	
		Unchanged	1	
Unfavourable group	3	Successful	2	(one had 8 children before operation)
		Unchanged	1	

In the following 6 cases, deafness was stated to be *unchanged* during pregnancy:

Favourable group	3	Improved	2	
		Unchanged	1	
Borderline group	3	Improved	2	(1 case—3 children before and 2 after operation. 1 daughter, aged 12, already has otosclerosis)
		Unchanged	1	

Information regarding *pregnancy after operation* is available in only 2 cases. In one favourable and successful case, hearing was not affected. The second was a "borderline" case in which post-operative hearing was worse and in which further deterioration occurred in both ears in association with each of 2 post-operative pregnancies.

"Flamingo Pink" Shimmer (Schwartze's Sign)

This was present in 10 cases in which the post-operative results are as follows:

Pre-operative Group		Result		Opposite ear		Operation note
Favourable	7	Successful	5	Unchanged Worse	4 2	—vascular enchondral bone—1
		Improved	1	?		
		Worse	1	Unchanged		vascular enchondral bone
Borderline	1	Worse	1	Unchanged		
Unfavourable	2	Unchanged	2	Unchanged	2	

A fistula sign remained present in every case.

Acute Otitis Media

Post-operative acute otitis media occurred in 5 cases at periods from 6 to 18 months. Four were successful results and no hearing impairment has resulted. One had hearing at pre-operative level with a positive fistula sign and no further deterioration has taken place.

Post-operative Hearing in the Non-operated Ear

In no case was significant hearing improvement found at any time in the non-operated ear. It remained unchanged in the majority. Post-operative deterioration occurred, however, in 29 cases, results in the fenestrated ear being as follows:

Pre-operative Group	Number	Successful	Improved	Unchanged	Worse
1. Favourable ..	19	15	1	1	2
2. Borderline ..	7	1	5	0	1
3. Unfavourable	3	0	1	1	1
	29	16	7	2	4

Fistula Sign

In three cases a fistula sign was never at any time found present:

1 in Favourable group; hearing unchanged. (Excessive bleeding during second stage of operation.)

1 in Borderline group: hearing worse.

1 in Unfavourable group: hearing unchanged. (Excessive bleeding during second stage of operation.)

Vertigo

In no case in the present series was vertigo a pre-operative symptom.

Phenobarbitone was given during the immediate post-operative period in the majority of cases where vertigo was other than minimal and observers unanimously consider that it is preferable to Avomine or Dramamine at that stage.

During the latter three years normal saline, formerly used for continuous irrigation during actual fenestration, has been replaced by a solution, supposedly isotonic with endolymph (Lamp 1952). No very definite reduction in the degree of post-operative vertigo and nystagmus has been observed, nor has a consistently earlier improvement in hearing resulted following its employment.

Head exercises, commencing 2-3 days after operation, became almost a routine. It is considered that they materially expedite rehabilitation, especially in older patients.

Late, post-operative vertigo has been encountered in 3 cases, all of which have a successful hearing result. In one case this persisted for a year, during which a very violent fistula sign was present. Transient vertigo on blowing the nose has persisted for more than 2 years in another. The third developed typical Meniere attacks 6 months after operation These have continued for more than 5 years with remissions and so far have failed to respond to any conservative treatment She continues to have a very successful hearing result with no deterioration whatsoever but it is noteworthy that immediately following attacks of vertigo audiograms have repeatedly shown a temporary loss of as much as 60 db Can it be that decompression of the labyrinth has saved Corti's organ from pressure atrophy? The fistula sign remains somewhat violent

Taste

Subjective unilateral loss of taste was complained of in one case in which the stapes was fractured the stapedius tendon being divided in order to effect removal of the fractured crura The chorda tympani nerve was not severed but was probably bruised The hearing result was satisfactory

The *chorda tympani nerve was deliberately divided* in 6 cases none of which had any subjective complaint or any other noticeable effect

Facial Paresis

Facial paresis of varying degrees of severity short of complete paralysis occurred after operation in 6 cases all of which recovered completely within periods of 2 to 12 weeks

TEMPORARY FACIAL PARALYSIS

	Day of onset	Duration —weeks	Pre operative Group	Hearing result	Observations
1	2nd	4	Unfavourable	Improved	Bone dehiscence of horizontal Fallopian canal
2	3rd	1?	Borderline	Unchanged	Post operative vertigo + +
3	10th	3	Favourable	Successful	Drip failed—? heating
4	11th	2	Favourable	Successful	—
5	11th		Favourable	Successful	Ultimate fenestra below level of facial nerve
6	1st stage of operation	12	Borderline	Successful	Facial twitch when cutting off malleus head

In Case 6 immediate microscopic inspection of the Fallopian canal revealed no abnormality It was thought that the thin bony wall of the horizontal Fallopian canal was momentarily indented by the malleus cutter thus squeezing the nerve Paralysis was immediate and severe but not complete The patient was able to resume work as an express engine driver at the end of three months

While the *duration of paresis* is in some measure related to the degree of its severity experience suggests that a severe paresis occurring relatively late is likely to recover more quickly than a relatively slight paresis which appears in the earlier post-operative period.

Anæsthesia

General anæsthesia has been employed in all cases, but various agents have been in use during different periods. *Oro-tracheal intubation* has invariably been carried out.

In assessing the relative value of different procedures, due consideration has been given to the individual preference of the anæsthetist. Previous to the employment of methods of blood pressure control, continuous *intravenous pentothal sodium* with adequate premedication gave the most consistent satisfaction (17 cases) second place being taken by *nitrous oxide/trilene* following intravenous pentothal sodium induction (28 cases). *Avertin* followed by nitrous oxide, favoured by one anæsthetist, was too frequently associated with troublesome hæmorrhage (24 cases). Experience of intravenous pentothal sodium combined with hypotension produced by *high spinal block* (Hall and Millar, 1950) has been limited to 15 cases. The fact that several different anæsthetists took part in this series may account for the undue degree of anxiety which this procedure afforded the operator. The *anæsthetic of choice* has been pentothal sodium induction followed by nitrous oxide/trilene, combined with production of hypotension with Hexamethonium *when required* (43 cases). In an appreciable number of cases the latter is never called for. When employed it is usually reserved for bleeding during the second stage. Only occasionally is it called for during the first stage. When used throughout the operation it has on occasions been found not possible to maintain an adequate reduction of pressure over the required period, even up to 1½ hours. If given selectively, however, this shortcoming can nearly always be avoided.

The Mastoid

Mastoid air cell development was regarded as normal in the great majority of cases, but cells were more or less absent in 16.

No rigid policy has been pursued, or indeed seemed practicable, concerning the extent of bone exposure. This has varied from the minimal required for access to complete exenteration of air cells, and has been determined largely by individual anatomical variations. Whenever possible, a well-shaped cavity of moderate size has been aimed at.

A secondary operation for exenteration of infected tip cells was required in one case, 2½ years after fenestration. Pre-operatively this was an unfavourable case in which hearing was unchanged throughout.

A two-stage operation was carried out only once and this was determined by very excessive bleeding with consequent detachment of the tympano-meatal flap, which was replaced at the end of the first-stage operation The second stage was carried out 4 months later, when the enchondral bone was found to be vascular and endosteal bleeding occurred The case was an "unfavourable" one and ultimate hearing was unchanged with a persisting positive fistula sign

The Stapes

Attempts were made to assess the absence or presence of mobility of the stapes in the oval window and assessment has been recorded in 85 cases It was regarded as being completely fixed in 63, of impaired mobility or "stiff" in 10 and quite mobile in 12 cases The following table of comparative results fails to show any significant features

Stapes	Pre operative Group	Number of operations	Successful	Improved	Unchanged	Worse
Fixed	Favourable Borderline Unfavourable	45 11 7	35*† 1 1	5 6 3	3 3 2	2 1 1
	All Groups	63	37	14	8	4
Impaired mobility	Favourable Borderline Unfavourable	7 2 1	4 2 0	0 0 0	3 0 1	0 0 0
	All Groups	10	6	0	4	0
Mobile	Favourable Borderline Unfavourable	8 3 1	7† 0 0	0 2 0	0 1 1	1 0 0
	All Groups	12	7	2	2	1

* 1 case—ankylosis of malleus incus
† 1 case—short process of incus ankylosed to aditus

Accidents During Operation

The *tympanic membrane was injured* with resulting perforation in 5 cases, all of which eventually healed This occurrence was essentially associated with troublesome bleeding

Pre operative Group	Number of operations	Hearing result		Healing time —weeks
Favourable	1	Successful		20
Borderline	1	Improved		7
Unfavourable	3	Unchanged	2	i 9 ii ?
		Worse	1	8

The *tympano meatal flap became detached* and was re-inserted after completing the operation *in one stage* in 3 cases Results

1 Favourable case: hearing unchanged—persistent fistula sign.
1 Borderline case: hearing improved—persistent fistula sign.
1 Unfavourable case: hearing satisfactory.

The *membranous canal was severely damaged* in 3 cases, all of which have satisfactory hearing. The broken tip of an excavator disappeared into the ampulla in one (unfavourable) case. The ultimate hearing remained unchanged and the fistula sign persists. The patient has shown no other unusual features.

An *unduly small fenestra* was made in 7 cases, 4 of which have been successful, 2 improved (1 Borderline, 1 Unfavourable) and 1 unchanged (Borderline with fistula sign). In no case has closure occurred.

Appreciable *bleeding from enchondral bone* was encountered in 5 cases, 3 of which had the "Flamingo Pink" shimmer. Hearing is satisfactory in 2, unchanged in 1 (unfavourable), and worse in 2 (1 favourable, 1 borderline). A fistula sign persists in all.

Adhesions between bony and membranous canal were encountered in 3 cases. No damage resulted in one (successful), the membranous canal was severely bruised in the second (successful) and became completely detached along with the bony canal "lid" in the third, so that the fenestra presented a cadaver-like appearance. Improved hearing has been maintained, with a feeble fistula sign (Borderline case).

Bleeding from the fenestral endosteal margin occurred in 19 cases. In no case was the flap placed over the fenestra till such bleeding had entirely stopped. Results are relatively poor:

Pre-operative Group	Number of Operations	Successful	Improved	Unchanged	Worse
Favourable . .	10	7	2	1 (Fistula +)	0
Borderline . .	5	0	4	1 (+)	0
Unfavourable . .	4	0	1	1 (+)	2 (+)
All Groups . .	19	7	7	3	2

Residual endosteal fragments which, for various reasons it was considered imprudent to attempt to remove, were recorded in 5 cases in which results are also poor:

Pre-operative Group	Number of Operations	Result	Fistula sign
Favourable . .	1	Unchanged	never +
Borderline . .	3	1 Successful 1 Improved 1 Worse	—never +
Unfavourable .	1	Unchanged	never +

"Favourable" Cases Not Successful After Two Years

As a corollary to the previous paragraph, 19 operations under this category are reviewed with the object of trying to ascertain the unfavourable features to which failure might be attributed

(*a*) *Four cases initially successful which relapsed to "Improved" hearing within 6 months*

- i "Flamingo pink" shimmer Fenestral endosteal bleeding at operation
- ii History of acute otitis media previous to operation Stapes fixed Persistent fistula sign
- iii Bony closure of fenestra (Result of revision operation not yet known)
- iv Straightforward operation Stapes fixed Persistent fistula sign

(*b*) *Three cases initially successful which relapsed to the pre-operative hearing level within 6 months*

- i Fenestral endosteal bleeding at operation Stapes "stiff ' Persistent fistula sign
- ii Excessive bleeding during operation—tympano-meatal flap detached and replaced in one-stage operation Stapes fixed Persistent fistula sign
- iii Troublesome bleeding during operation Stapes "stiff" Persistent fistula sign Exploratory revision operation—fenestra patent (see under Revision Operations)

(*c*) *Five cases never successful, but with "Improved" hearing*

- i Fenestral cartilage "stopple" (see under Revision operations)
- ii Fenestral endosteal bleeding at operation Persistent fistula sign
- iii Ankylosis between malleus and incus Stapes fixed Persistent fistula sign
- iv Straightforward operation Stapes fixed Persistent fistula sign
- v Stapes mobile Persistent fistula sign Long-continued attic mucoid discharge associated with nasal allergy Resolution after A C T H Probably chronic adhesive process

(*d*) *Three cases never successful and with hearing remaining at pre-operative level*

- i Troublesome bleeding during operation Fistula sign never present
- ii and iii Straightforward operation Stapes fixed Persistent fistula sign

(*e*) *Four cases never successful and with hearing worse than before operation*

- i Stapes fixed Severe post-operative vertigo Persistent fistula sign

(Previous operation on the other ear, performed by another surgeon, was also a failure.)

ii. Troublesome bleeding during operation. Stapes fixed. Fistula sign never present.

iii. "Flamingo pink" shimmer. Vascular endochontral bone. Stapes mobile. Persistent fistula sign.

Healing of Cavity

Duration of stay in hospital: In addition to the usual determining factors, the distance from hospital of the patient's home frequently necessitated prolonging the stay, particularly when the patient lived too far away to be able to attend at frequent short intervals.

In the series of cases under review, the majority were in hospital for three weeks, the minimum time being two weeks and the maximum eight, giving *an average of three and a half* weeks. The average duration of stay of ungrafted cases was 3·5 weeks, while the average period for those in which a primary Thiersch skin graft was applied was 3·3 weeks.

Healing time: A comparison has been made between *non-grafted* and *Thiersch skin-grafted* operations:

	Number of Operations	Healing time: weeks		
		Minimum	Maximum	Average
Not grafted	91	3	32	*10·8*
Primary Thiersch grafts:				
Successful	28	3	24	*8·8*
Failed	3	6	36	—
Secondary Thiersch grafts (1 week after operation) ..	2	5	12	—

With the exception of the two secondarily grafted, *none* of these cases received penicillin during the immediate post-operative period.

It is thus shown that successful primary Thiersch grafting has not effected any reduction in the duration of stay in hospital, but has effected some reduction in the healing time.

Penicillin has been given as early post-operative routine only in 29 cases, in which the healing time varied from 4 to 32 weeks. The average time being 10·3 weeks compared with 10·8 weeks in the group of 91 non-grafted cases which did *not* receive penicillin.

A comparison of hearing results in these two groups, in cases in which *healing was delayed for 20 to 32 weeks*, hardly seems significant:

Penicillin *not* given—5 cases—successful 3. Unchanged 2 (fistula +).

Penicillin given—4 cases—all successful.

Late Recurrent Infection

Late recurrent infection of the mastoid cavity has proved to be the most troublesome post-operative feature In some cases this is due to *neglect* on the part of patients who fail to attend for toilet at regular intervals In 9 cases, however, operation afforded inadequate closure of the *anterior attic* by the tympano-meatal flap (generally associated with troublesome bleeding during its preparation) Analysis of these 9 cases shows that all but one had recurring discharge, commonly in association with colds, but there is no evidence to suggest that hearing was prejudiced

Pre operative Group	Number of Operations	Hearing result	Primary healing—weeks	Infection
Favourable	6	All successful	5—16	nil 1 recurring 5
Borderline	3	Improved 2 Worse 1	8—16	all recurring

The *incidence* of recurring infection has been compared in three groups of cases

	Number of Operations	Average healing time—weeks	Recurring infection
1 *No* penicillin	91	10 8	23%
2 Penicillin post operation	29	10 3	28%
3 Successful primary Thiersch graft (*no* penicillin)	28	8 8	32%

It appears that the *lowest* incidence of recurring infection occurred in the no penicillin/no graft group and the *highest* in the grafted cases

Shambaugh and Derlacki (1956) found that primary Thiersch grafts did not shorten the time of healing and that the thin epidermis dequamates excessively, breaking down frequently with recurrence of discharge They advise against its use They also found that covering the fenestra with free Thiersch graft instead of the usual flap offers no advantage

Size of external meatal opening experience suggests that a relatively large meatal opening, though perhaps sometimes regarded as being somewhat conspicuous, not only expedites primary healing but also reduces the liability to recurring infections of the cavity

Infection and Hearing Results

Details are available in 85 out of the total of 92 operations in the "Favourable" pre-operative group which allow a comparison of hearing results to be made between (*a*) cavities healed within approximately

12 weeks with no recurring infection and (*b*) cavities in which the primary healing time has been prolonged (12-32 weeks), or in which recurring infection has occurred after primary healing:

Hearing result	Normal healing	Prolonged healing time or recurring infection
Successful	36=*82*%	33=*81*%
Improved	3	4
Unchanged	3 (2 with fistula +)	3 (all with fistula +)
Worse	2 (fistula +)	1 (fistula —)
	44	41

The similarity of percentage successful results in the two groups suggests that prolonged healing time and recurring infection have not prejudiced late hearing results.

Early secondary infection with *Ps. pyocyaneus* occurred in 2 cases attributed to cross-infection. Hearing results were as follows:

1 Favourable case—successful.

1 Borderline case—improved.

Bacteriology

In *recurring infections* the prevailing organisms found were *Staphylococcus aureus*—penicillin-resistant,—penicillin-sensitive, *Ps. pyocyaneus* and *B. Proteus,* in that order of frequency. One cavity was infected with *Aspergillus niger.*

Revision Operations

Eight revision operations have been performed out of the series of 205 fenestrations. In no instance has more than one revision so far been indicated. Five cases were females and three males.

The *ages* at the time of the original operation were from 17 to 37, the average being 26, which is appreciably lower than the average of 35 for the whole series. Seven were in the "favourable" pre-operative group and one in the "borderline" group.

Primary hearing results: 5 operations were initially successful, 2 were appreciably improved and one remained unchanged after the original operation.

Time of hearing relapse: 4 successes and one improved case relapsed to pre-operative level *within 6 months.* One successful and one improved ("stopple") case relapsed within one year.

The *fistula sign* was initially present in 7 cases. It had ceased to be present within 6 months in 5 cases and within a year in one ("stopple"). It was never present in one case and continued to be present in another.

The *time of revision operation* varied from 9 months to 5 years, the average being 2½ years

Findings at revision operation bony closure was complete in 5 cases and almost complete in one In one case (of only 2) in which a cartilage "stopple" had been inserted, it was found to be overgrown by bone The fenestra was fully open in one

Histological examination of the "closure" was carried out in 4 cases In 2 instances the finding was "Bone recently formed by metaplasia from fibrous tissue" and in another "Compact bone of recent formation" The removed "stopple" showed metaplastic bone formation

Hearing results one case is too recent to ascertain the result and one is so far successful after 3 months 4 remain successful after periods of 4, 2, 2, and 1 years The "stopple" case, after initial success at 3 months, relapsed to an improved level which has been maintained for 2 years One case remained unchanged In 2 so far successful cases the hearing after revision was better than its best after the original operation

The fistula sign did not reappear after revision of the "stopple" case It did reappear in the remaining 6 on record, but in 2, in which hearing is satisfactory, it did not reappear till more than 6 months after revision In one of these it had never been present after the original operation

Two cases merit special notice

1 The "stopple" case, in which the stopple had passed too deeply into the canal and its edges had become overgrown with bone

2 A male aged 18 at the time of his original operation which was uneventful in spite of somewhat excessive bleeding and who was in the " favourable" pre-operative group The stapes was found to be "stiff" Initial satisfactory hearing relapsed to the pre-operative level within 6 months, but the fistula sign remained well-marked In spite of this, re-exploration was carried out after 3 years At operation the covering flap was found to be elevated off the fenestra which appeared as though newly made The flap was firmly replaced and indented according to Lempert Hearing, however, has remained at its pre-revision level during the ensuing year, while the fistula sign remains active

The table on p 116 summarizes the foregoing account of *Revision operations*

Summary

1 Of a series of 205 fenestration operations, follow-up of 139 (on 136 patients), carried out from 2 to 10 years ago, has been possible

2 Cases have been classified pre-operatively as Favourable, Borderline and Unfavourable and results have been reviewed accordingly Successful hearing results have been obtained in 79 per cent of "Favourable" cases

3 The possible prognostic significance of certain features is considered

Case No	Pre-operation Group	Result after Fenestration						Interval between Fenestration and Revision	Findings at Revision	Histology	Results after Revision					
		Hearing			Fistula sign						Hearing			Fistula sign		
		3/12	6/12	1 year +	3/12	6/12	1 year +				3/12	6/12	years	3/12	6/12	years
1/1	F	Imp	Imp	Pre	+	+	—	5 years	"Stopple"—too deep, edge overgrown by bone.	" Stopple" metaplastic bone formation	S	Imp	Imp 2	—	—	— 2
2/53	B	Imp	Pre		+	—		9 months	Complete bony closure	?	S	S	S 4	—	—	+ 1 and after
3/95	F	S	Pre	Pre	+	+	+	3 years	Flap elevated off Fenestra. Fenestra satisfactory.	?	Pre	Pre	Pre 1	+	+	+ 1
4/131	F	S	Pre	Pre	—	—	—	1 year	Complete bony closure and adhesions	Bone recently formed by metaplasia from fibrous tissue	S	S	S 2	—	—	+ 1 and after
5/132	F	S	Pre	Pre	+	—	—	2 years	Complete bony closure	?	S	S	S 2	+	+	+ 2
6/134	F	Pre	Pre	W	+	±	±	3 years	Almost complete bony closure Membraneous ampulla injured	Bone recently formed by metaplasia from fibrous tissue	S	?	?	+	?	
7/143	F	S	Pre	Pre	+	—	—	2 years	1st op Vascular enchondral bone. Revision complete closure with soft bone Membranous canal very atrophic	?	S	S	S 1	+	+	+ 1
8/158	F	S	S	Pre	±	±	—	2 years	Bony closure	Compact bone of recent formation		?			?	

F = Favourable
B = Borderline

S = Successful
Imp = Appreciable improvement

Pre = Pre operative
W = Worse.

+ = Present
— = Absent

4 Some post operative complications and experience of various general anæsthetic agents are discussed

5 Unusual operative findings, difficulties and accidents are recorded and the influence of these on results is examined

6 The process of healing and the value of skin grafting are examined Late recurrent infection has proved to be the most troublesome post-operative feature, but evidence does not suggest that the late hearing result is prejudiced thereby

7 Eight Revision operations are considered in detail The hearing result has so far been successful in 4 cases for periods of 4 2, 2 and 1 years One case is successful after 3 months, one, initially successful, relapsed after 3 months and one remains unchanged

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CIRCULATION OF LABYRINTHINE FLUIDS

By L. NAFTALIN and M. SPENCER HARRISON

CERTAIN features of the physiology and pathology of the labyrinth have been difficult to explain on the basis of present concepts of the circulation of the labyrinthine fluids. Indeed, the majority of authors do not speak specifically of a common circulation and it has been generally assumed that the fluids, perilymph and endolymph, are almost entirely independent; whilst again it has not been clearly stated that they are static, no full description of a dynamic flow has appeared. It is proposed here to examine the physiology of these fluids in the light of more recent research in an attempt to give a more integrated picture of the structure and function of the inner ear.

Our knowledge of the nature and function of the labyrinthine fluids until quite recently was based chiefly upon the anatomical data found in the well-known atlas of Retzius (1881). It was believed that the perilymph spaces, connected through the cochlear aqueduct with the cerebrospinal subarachnoid cistern, were in effect a diverticulum of the latter in which was immersed the closed sac of the membranous labyrinth, derived from the invaginated ectoderm of the otic vesicle and filled with fluid of its own, the endolymph.

A more complete picture has gradually evolved, Waltner (1948) and many others have described a limiting membrane, from the arachnoidal web, which bars the labyrinthine opening of the aqueduct. The source and circulation of the endolymph were described by Shambaugh (1908) who considered it was secreted by the stria vascularis in the outer wall of the scala media of the cochlea. Guild (1927) carrying the investigations further injected solutions into the guinea-pig's endolymph system and considered that a current flowed from the cochlear duct to the saccule and finally through the wall of the saccus endolymphaticus.

The perilymph is not difficult to obtain in sufficient quantities to analyse, but much greater technical difficulties have been encountered in obtaining samples of endolymph which in addition must be small and accordingly particularly difficult to examine chemically. Measurements of the osmotic pressure of the labyrinthine fluids were reported in 1940 by Aldred, Hallpike and Ledoux, followed by the valuable studies of Ledoux (1941, 1943 and 1950) of the physical and chemical properties of these fluids. More recently a detailed account of modern microchemical measurements of the electrolyte contents of the labyrinthine fluids of the guinea-pig and cat have been provided by Smith (1954) and Citron, Exley and Hallpike (1956) (Tables I and II).

The fact that the protein content of the perilymph is higher than that of the cerebrospinal fluid has an obvious bearing upon current views on the interchange of these fluids that was thought to take place through the cochlear aqueduct and might suggest the existence of a semi-permeable barrier, so retaining the large protein molecules within the perilymph, while permitting interchange of the smaller electrolytes. The high protein content of the perilymph has, on the other hand, been explained as a stagnation phenomenon similar to that occurring in the subarachnoid space in association with the localized restrictions in the circulation of the cerebrospinal fluid with increase of its protein content. The high potassium and low sodium content of the endolymph are unique for extracellular fluids. This might seem to establish a biochemical similarity between the endolymph and other intracellular fluids elsewhere in the body and it has been supposed that the endolymph is produced by secretory activity of some specialized cells. Smith (1954) notes the difficulties of nerve impulse transmission along un medullated nerve fibres which traverse the tunnel of Corti's organ apparently immersed in the endolymph. Citron, Exley and Hallpike (1956) suggest that it is possible

TABLE I

Analysis of Labyrinthine Fluids in the Cat and Guinea Pig. Results of Previous Investigations as Given by Citron, Exley and Hallpike

Animal	Properties and Constituents	Spinal fluid	Perilymph	Endo lymph	Reference
Cat	Osmotic pressure (g NaCl/100 g H_2O)	1·017	1·046	1·058	Aldred, Hallpike and Ledoux (1940)
	Index of refraction (n_D22°C.)	1·33435	1·33495	1·33455	Ledoux (1941)
	pH	7·45	7·87	7·82	Ledoux (1943)
	Chloride (m equiv./l)	152	158	151	Ledoux (1950)
	CO_2 (vol./100 ml s.t.p.)	43·2	32·0	31·5	
	Protein (mg./100 g.)	31	268	118	
Guinea pig	Sodium (m equiv./l)	152	150	16	Smith, Lowry and Wu (1954)
	Potassium (m equiv./l)	4·2	4·8	144	
	Chloride (m equiv./l)	122	121·5	107	

that the fluid system within Corti's organ itself is an enclave separated from the endolymph space, containing fluid with biochemical properties which do not differ from those of extracellular fluids surrounding nerve fibres in other parts of the body, but it is difficult to conceive a separate fluid system within the organ of Corti, particularly as parts of the cell mass are likely to be surrounded by and bathed in endolymph from which, according to current theories, they derive their nutriment. In fact, Kolmer (1927) states that Corti's organ derives its oxygen and other requirements through the medium of the endolymph as no capillaries are present in this organ.

TABLE II.

ANALYSIS OF LABYRINTHINE FLUIDS IN THE GUINEA-PIG (CITRON, EXLEY AND HALLPIKE).

Constituent	Serum	Spinal fluid	Perilymph	Utricular endolymph
Sodium (m-equiv./l)	140 (4)	150 (12)	148 (12)	26 (8)
Potassium (m-equiv./l.)	4·5 (4)	4·0 (12)	5·0 (12)	142·0 (9)
Chloride (m-equiv./l.)	—	122 (9)	120 (9)	110 (3)
Protein (mg./100 g.)	—	20 (6)	75 (6)	25 (6)
Non-protein nitrogen (mg./100 g.)	—	21 (6)	20 (6)	21·5 (6)

ANALYSIS OF LABYRINTHINE FLUIDS IN THE CAT.

Constituent	Serum	Spinal fluid	Perilymph	Cochlear endolymph
Sodium (m-equiv./l.)	155 (12)	162 (12)	164 (12)	66 (12)*
Potassium (m-equiv./l).	6·7 (12)	5·9 (12)	6·0 (12)	117·0 (12)*
Chloride (m-equiv./l.)	—	150 (4)	150 (4)	—
Protein (mg./100 g.)	—	25 (5)	142 (5)	—
Non-protein nitrogen (mg./100 g.)	—	20 (5)	21 (5)	—

The *stria vascularis* is a compact band of specialized epithelial cells not seen elsewhere in the membranous labyrinth, enclosing an extensive capillary network. Together with the cells of the spiral prominence the stria forms the lateral wall of the cochlear duct. The endolymph surface is enclosed by marginal cells and the capillary network is in the epithelial layer. Catherine Smith (1957) has recently given a detailed account of the histology of the *stria vascularis*. Pease (1956) compares the intense infolding of the basal plasma membrane with that of a similar type of structure in the kidney tubules and the submaxillary gland. Palade (1956) observed a similarity between the marginal cells and macrophages, suggesting they may be a route for phagocytosis Cellular debris is rarely seen close to the stria vascularis (Smith 1957) and may be removed by these cells The vestibule contains no cell mass resembling the stria, but the cells of the spiral prominence are duplicated in this part of the labyrinth Belemer (1936) described vessels passing through the modiolus across the roof of the scala vestibuli and entering the stria at its upper end These vessels subsequently pursue a course of varying obliquity through the cell mass to emerge once more from its lower border into the substance of the spiral ligament, the vessels of the spiral prominence not being found in its epithelium The vessels of the stria are intimately related to the superficial cells and maintain their calibre in their passage through it

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* In many of these samples the Na+ and K+ concentrations suggested contamination with perilymph. Three samples, however, showed the high values for K given in the table.

The publications of Mygind and Derderding (1929, 1938, 1943) constituted the first approach to the biochemistry of the labyrinthine fluids They started with the premise that Menière's disease resembles glaucoma and is a hydrops of the labyrinth, originating they postulated, in a nutritional disturbance produced by capillary dysfunction with a resultant defective oxidation and increase in water-binding efficiency in the cells with subsequent intracellular œdema They succeeded in relieving many cases, but the treatment did not strike at the origin of the trouble, namely the cause of the localized accumulation of fluid in the labyrinths of patients with an otherwise normal metabolism

Furstenberg and his colleagues (1934, 1936) re investigated the problem and produced evidence which indicated that a retention of sodium salts was of greater pathological significance than a retention of water in the production of symptoms The concentration of electrolytes and the degree of hydration of the blood was not reported in any of these communications, and it was left to Talbott and Brown (1940) to show that in four patients during an attack the serum potassium was raised and the sodium was lowered, whereas in forty four patients between attacks the concentrations of sodium, potassium, calcium, chlorides, carbon dioxide, phosphate, protein and non-protein nitrogen were normal, and there was no change in the acid base equilibrium of the blood in Meniere's disease They administered sodium salts, including the bicarbonate, orally and parenterally without precipitating an attack They concluded that the idea must be abandoned that the symptoms were due to gross retention in the body of water or of sodium chloride, because, if such were so, changes —not in practice found—would be present in the relative concentrations of sodium, protein and water in the plasma This conclusion is, however, open to the criticism that it makes no allowance for the equilibrating changes which may take place between these plasma constituents and the total body pools Talbott and Brown (1940) explained the clinical benefits of their diet by stressing the importance of the potassium content of the blood, since it has been observed to vary in certain diseases in which the conduction of nerve impulses is disturbed Normally the proportions of sodium and potassium in the diet are equal, but a relative increase in the potassium is inevitable in Furstenberg's regime. Since this diet is effective, they investigated the combination of a normal diet supplemented with extra potassium (6-10 gm. equal, approximately, to 80-134 m. eq. potassium chloride daily), and found this an apparently successful method of therapy. Other authors, e.g. Walsh and Adson (1940) have recorded similar results.

No discussion on the movements of body sodium and potassium, and the factors controlling the homeostasis of these electrolytes, would be complete without mention of the part played by aldosterone. Although it is known that aldosterone is the principal adrenal steroid responsible for sodium conservation, and, to a lesser extent, potassium excretion, it is not known what controls the rate of secretion of aldosterone (*Lancet*, 1957).

A description of the effects of changes in the dietary loading of electrolytes on the aldosterone output in the urine (and presumably, therefore, also on the rate of secretion of aldosterone by the adrenals into the blood) has been given by Johnson *et al.* (1957). From their own work, and that of others, they draw several conclusions which may be listed as follows:

Normal subjects on an intake of normal diet except for:

(1) Sodium depletion, have an increased output of urinary aldosterone.
(2) Sodium loading, have a decreased output of urinary aldosterone.
(3) Potassium loading, have an increased output of urinary aldosterone.
(4) Potassium depletion, have a marked decrease in the output of urinary aldosterone.
(5) Potassium depletion + sodium deprivation, have a delayed increase in the output of urinary aldosterone, the increase not being as great as that observed during sodium deprivation alone.
(6) Sodium restriction being maintained, but restoration of potassium to the diet, have a further slight to moderate increase in urinary aldosterone over the output recorded for condition (5).

These authors found that "relatively large changes in aldosterone output were observed without striking alterations in the concentration of plasma electrolytes, but the decreases or increases in aldosterone excretion after each change in the diet were in a direction favouring homeostasis of body sodium and potassium".

These observations and conclusions would appear to support the view that relatively small fluctuations of dietary intake of electrolytes, or the minor degree of sodium deficit produced by a "low salt" diet, would not by themselves be sufficient to alter the body balance of sodium and potassium to any significant extent.

Aldosterone exerts its principal effect, from the point of view of sodium conservation, on the renal tubule. The renal tubular cell has to perform enough work to transfer potassium from the plasma to the urine and to move sodium in the opposite direction, both transfers taking place against concentration gradients. This work is done with a minimum of energy output by means of an ion exchange mechanism, and it may be that one effect of aldosterone is to facilitate this work The situation in the endolymphatic system could be of a similar nature Elsewhere in the body extracellular secretions are usually of high sodium content, i e they are modifications of simple plasma transudates, and osmotic and ion transfer work is kept to a minimum Both endolymph and urine in certain circumstances have a high potassium content and a low, or relatively low, sodium concentration So much is this the case with endolymph that it has been described as having an "intracellular" character It is here suggested that it may be simpler to regard endolymph as starting out as a plasma transudate, and that selective reabsorption and secretion by the stria vascularis alters the sodium and potassium concentrations This hypothesis has the advantage of requiring no special work, or change, or new secretion, as regards non-protein nitrogen, calcium and hexosamine, to produce endolymph from perilymph There is the further advantage that it is simpler to conceive of an active circulation by this hypothesis So long as endolymph is regarded as a secretion into a closed system there is difficulty in describing an adequate circulation in the absence of a drainage point or canal, and no such exit has yet been noted If, now, circulation is assumed to start from a plasma transudate, having the hydrostatic pressure of the general circulation to provide the energy, and this transudate is, or becomes, perilymph and passes through Reissner's membrane to become endolymph, it is possible to visualize a sufficiently dynamic flow as to bring oxygen in physical solution in adequate quantities to the cells of the organ of Corti The special character of the endolymph would be maintained by selective absorption and secretion by the stria vascularis

A number of observations and experiments will be cited in support of this concept, but it is strongly emphasized that the starting point of any theory of function, or of abnormality, of the endolymphatic system must be the description of how oxygen reaches the organ of Corti Perlman and Kimura (1957) state that within seconds after respiratory oxygen is replaced by nitrogen, or after general blood flow is impaired, functional changes are demonstrable in the labyrinth It is reasonable to suppose from this short time relationship, that the cells to be first affected by anoxia will be those having a specialized function but furthest away from a good capillary network Since histologically it is difficult to demonstrate any direct blood supply to the organ of Corti, and since, from their situation and contiguity with the nerve endings of the auditory nerve, it may be assumed that Hensen's, Deiter's and the hair cells have an active metabolism and therefore require a steady oxygen supply, it seems fair to conclude that it is in the cells of the organ of Corti that the impaired functional changes occur in a matter of seconds-when oxygen flow fails. This conclusion can be drawn also from the evidence provided by experiments concerning the electrical phenomena of the cochlea. The subject has been reviewed recently by Davis (1957). In this review the evidence is summarized showing that one of the electrical responses to acoustic stimulation (called the cochlear microphonic) is related to the displacement of the tectorial membrane relative to the reticular lamina and is dependent on the integrity of the hair cells. The cochlear microphonic is closely dependent on an adequate oxygen supply. The action potential response (of the acoustic nerve impulses) is even more closely dependent on oxygen supply than is the cochlear microphonic. "The very prompt failure of the action potential and of the cochlear microphonic (when oxygen tension falls) argues for a high metabolic rate of the nerves and sensory cells and the absence of any significant storage mechanism for oxygen" (Davis, 1957). The first requirement therefore of any theory of formation and circulation of endolymph must be a description of the direction of the oxygen gradient.

Formation of endolymph may occur in one of several structures. The stria vascularis is assumed by most authors to be the source of endolymph, but, as has been already remarked, no drainage canal has been described to permit of a circulation starting from the stria vascularis, and a reabsorption through some membrane must therefore be postulated. Citron and Exley (1957) have pointed out that active transport across Reissner's membrane would seem to be necessary in any theory, but this structure is not so situated or constructed as to make it satisfactory, either as a source or exit of a circulation. It could, nevertheless, effect the changes in concentrations of sodium and potassium to transform endolymph into perilymph. Perilymph would then drain to the cerebrospinal fluid, the high protein content of the former being diluted by the larger volume of the latter. Drainage of perilymph may also occur along lymphatics, and Kley (1951) presents evidence suggesting that perineural flow and direct transudation of perilymph into the venous system are further possibilities.

There are serious objections to the theory of a circulation beginning at the stria vascularis and passing through the stages of endolymph and perilymph. The least serious is the difference in protein concentrations. To make endolymph into perilymph Reissner's membrane would have to add protein. A more serious objection is the question of oxygenation. If the stria vascularis is to be the site of secretion of endolymph it is necessary to suppose that oxygen is passing through three layers of actively metabolizing cells, i.e. cells having a large oxygen consumption. If, on the other hand, the stria vascularis is to be considered relatively inactive metabolically and allowing a plasma transudate rich in dissolved oxygen to pass, then we have the anomalous position that Reissner's membrane, a thin endothelial structure without a very abundant blood supply, has all the work of ionic change to perform while the stria vascularis acts only as a mechanical filter though it possesses a rich capillary network underlying cells whose micro-anatomical structure resembles more the renal tubular cell than the glomerulus (Smith, 1957, Pease, 1956, Engstrom, 1955) An important point in connection with oxygen supply is made by Perlman and Kimura (1957) who found, in experiments using guinea-pigs that anoxia of the cochlea and labyrinth produced by obstruction of the inferior cochlear vein, led to profound disturbance of vestibular function with nystagmus, head and body torsion and circling movements The animals were sacrificed at intervals after production of the lesion and the structures preserved by intravital perfusion of fixative Histological examination showed that the stria was particularly susceptible, often being the first to show signs of damage Dilatation of strial capillaries and œdema of the epithelium were noted one hour after venous obstruction Within a few hours hæmorrhage into the epithelium and into the scala media was noted Disintegration of the epithelial layer of the stria soon followed in some (animals) On the other hand within 24 hours after venous obstruction loss of outer hair cells was clearly evident, but the inner hair cells were more resistant and degenerative changes appeared after two days This description, and examination of a photomicrograph of a preparation from one of the experiments quoted, indicate that the organ of Corti had survived several hours at least after disintegration of the stria vascularis had begun This conclusion argues strongly that oxygen was reaching the organ of Corti from a source other than the stria vascularis It is worth emphasizing that in this case the organ of Corti survived the destruction of the stria while a characteristic of Menière's disease is that the stria remains intact while the organ of Corti degenerates It is difficult to escape the conclusion that the organ of Corti is not dependent on the stria vascularis for its oxygen supply and other nutriment

Perhaps the most serious objection to the theory that the stria vascularis is the source of endolymph is that raised by a consideration of ion transport energetics In Fig A there is diagrammatically represented the ionic situation in the case of a flow from plasma through the stria vascularis to the scala media, from there through Reissner s membrane to the perilymphatic space If the stria vascularis has actively to secrete K^+ and withhold Na^+ then it must do so, in this case without the use of an ion exchange mechanism Since the flow is away from the stria vascularis no Na^+ can reach the endolymphatic surface of the strial cells to permit of an equimolar exchange of sodium and potassium with minimum energy expenditure If a local transudation were occurring to provide sodium for reabsorption, the description of a circulation starting from the stria vascularis would no longer hold, since reabsorption would entail something of a reverse flow, or alternatively the correct description would be that of a transudate at the stria with the ionic work being done by Reissner s membrane because sodium in the strial transudate would immediately be distributed throughout the endolymphatic space.

The other cation exchange which could occur to minimize energy expenditure is that of K^+ for H^+. One must assume a blocking of Na^+ transport at the basal layer of the stria vascularis; the upper layers then have carbonic anhydrase mechanism for forming and dissociating carbonic acid, thus providing ionized hydrogen. H^+ could then exchange for K^+ and potassium bicarbonate would be secreted. Although the pH of endolymph is somewhat higher than that of plasma and this elevated pH could be explained by the secretion of potassium bicarbonate, the data of chemical analysis of the endolymph (Citron and Exley, 1957) do not support this idea. The endolymph has a low carbon dioxide content (Ledoux, 1950) and the exchange theory involving bicarbonate would require a high one. Further the chloride content of endolymph is high, i.e. of the same order as cerebrospinal fluid and perilymph, and yet another ion exchange mechanism would be necessary to explain chloride transport after separation from sodium in the basal layer of the stria vascularis. The chloride content is best explained by assuming the formation of a plasma transudate, but as pointed out above, the formation of transudate only by the stria leaving the active ionic transport work to be done by Reissner's membrane is in contradiction to their obvious histological structure.

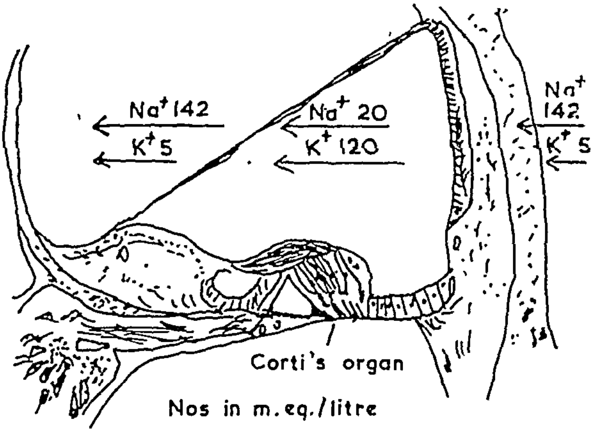

FIG. A

Fig. B presents a diagram of the ionic situation in the alternative case, i.e. the hypothesis suggested in this paper, of flow proceeding from perilymph through Reissner's membrane to endolymph, the stria vascularis acting as a selective absorbing site. In this case Reissner's membrane has no external ionic work to do at all except to prevent the flow of K^+ from the scala media to the scala vestibuli The reduction of protein concentration to produce endolymph from perilymph may be ascribed to membrane impermeability to large molecules The stria vascularis, on this hypothesis, is presented in the first instance with a high sodium low potassium fluid, from which it extracts the sodium and exchanges potassium for it by an ion exchange system analogous to that of the renal tubular cell, using a minimum of energy Since potassium is not to pass Reissner's membrane except by a relatively slow equimolar exchange for sodium, i e at a rate suitable to meet the needs of the stria vascularis, potassium concentration in the scala media can build up until the required concentration is reached A numerical example will illustrate this point if, let us say, a litre of plasma flows through the stria vascularis per unit of time, and the extraction of K^- from the plasma is 40 per cent efficient 2 m eq of K will be exchanged for Na^+ per unit time If the leakage rate across Reissner s membrane is 1 m eq per unit time (the leakage rate being thought of as a function of membrane thickness) then 1 m eq K^+ will have been effectively transferred from the litre of plasma to the volume of the endolymph The rise in concentration of K^+ in the endolymph will, therefore be dependent on the ratio of the volume of endolymph to the plasma flow (corrected for extraction efficiency) through the stria vascularis per unit of time and equilibrium will be determined by the structural characteristics of Reissner s membrane It should be noted that this hypothesis provides for the ionic work which has to be done against gradients to be done by the structure capable of doing it, viz the stria vascularis, while the equimolar exchange of Na^+ for K^+ through

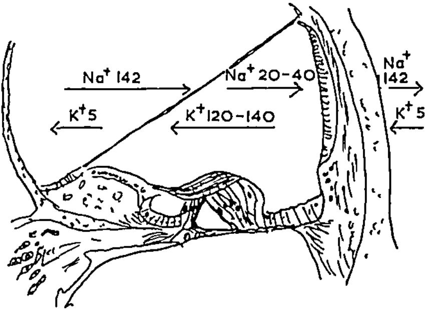

Fig B

Reissner's membrane takes place in the direction of the respective concentration gradients and hence requires no work on the part of that membrane. Osmotic pressure on both sides of Reissner's membrane is the same. No explanation is required for the concentrations of chloride, calcium, non-protein nitrogen and hexosamine since no change occurs in their concentrations, but it may be pointed out that this hypothesis shows an economy in the postulated number of secreting surfaces over other theories which require separate mechanisms to produce perilymph and endolymph, which have so many features in common.

If endolymph comes from perilymph it is necessary to enquire into the source, or sources, of perilymph. Kley (1951) performed experiments on guinea-pigs in which he injected fluorescein into the blood stream after blocking the cochlear aqueduct to prevent access of cerebrospinal fluid. Fluorescence nevertheless appeared in the perilymph simultaneously with its appearance in the aqueous humour of the eye. He suggested that perilymph was formed by ultrafiltration from the abundant vascularization of the perilymphatic space. In the human subject this source may be of greater importance than in animals since the cochlear aqueduct in man is much less permeable than in animals. Altmann and Waltner (1950) working with rabbits, cats and monkeys, and using the method of Weed (1914) of injecting iron salts into the subarachnoid space, sacrificing the animals after several minutes and determining the distribution of the iron salts by the Prussian blue reaction, came to the conclusion that, although a process of diffusion carried the salts from the cerebrospinal fluid through the cochlear aqueduct to the perilymphatic spaces, no active fluid currents appeared to exist in either direction. Their evidence suggests that the main part of the perilymphatic fluid is formed in the perilymphatic space, i.e. in agreement with the work of Kley mentioned above. Notable support of this view is obtained from a consideration of the protein concentrations of cerebrospinal fluid, perilymph and endolymph. The protein concentration of perilymph is four or more times higher than that of cerebrospinal fluid and also much higher than that of endolymph. Perilymph could thus not be formed from either cerebrospinal fluid or endolymph without further secretion, or alternatively reabsorption of all constituents except protein.

The evidence cited in the last paragraph provides a source of perilymph given the hypothesis that circulation in the fluids of the inner ear proceeds from perilymph to endolymph. That evidence suggests that perilymph is a plasma transudate formed on the scala vestibuli side of Reissner's membrane and, probably, also in the scala tympani on the lower side of the basilar membrane. This description permits the postulate to be made of an oxygen rich fluid having only a short distance to go to reach the organ of Corti, traversing only a membrane (either the basilar or Reissner's) which is doing little or no external work. A similar conclusion is reached by

Davis (1957) from the changes in electrical potentials when Ringer's or a Tyrode solution, modified to contain a high potassium and low sodium concentration, are substituted for endo- or perilymph He writes " the nutriment of the hair cells comes not from the endolymph but from the scala tympani (through the basilar membrane)"

This hypothesis thus takes account of the oxygen supply, requires a minimum number of exudative and absorptive surfaces, permits the description of a dynamic circulation using the hydrostatic pressure of the general circulation as a starting point and requires the minimum output of osmotic and ion transfer energy by the membrane cells concerned, allocating that work in accordance with their anatomical structure, and in accordance with the known ion concentration gradients

Certain objections to this theory must be considered Some experimental results (Guild, 1927, Altmann and Waltner, 1950) have been interpreted as meaning that injected granular material could be found in various parts of the scala media but seldom on the surface of the stria vascularis (Smith, 1957) Also cellular debris dislodged by acoustic trauma from the basilar membrane is rarely seen near the stria vascularis (Smith and Covell, quoted in Smith, 1957) Their interpretation is that the endolymph is formed by the stria vascularis and washes away the granules Altmann and Waltner did not inject granular material but rather soluble iron salts which were later precipitated as Prussian blue in the sites to which the salts had been diffused or had been carried Thus those authors' results, so far from suggesting that a fluid outpouring was washing away the granules from the stria vascularis, on the contrary might suggest a rapid resorption of iron salts through the stria They write, "Since salts are only resorbed in dissolved form together with fluids, fluid movements must exist, directed in the scalae towards the lower and upper parts of the spiral ligament, and in the cochlear duct towards the external spiral sulcus, the spiral ligament and the crista spiralis" However, they add that the fact that in their experiments the iron salts moved from peri- into the endolymphatic space simply demonstrates the high degree of permeability of the membranes and should not be taken as an indication that this is the prevailing direction of movement of dissolved substances under physiological conditions "Normally", they write, "it is more likely to be from the endolymph to the perilymph" Whatever the final interpretation of this work, it does not appear to signify such a flow of endolymph from the stria as to sweep the soluble iron salts (not granules) away

The problem of the accumulation of debris in other parts of the scala media, and elsewhere in the membranous labyrinth, but not over the stria vascularis, must be examined Waltner (1954) in his study of perilymph formation and composition found that on removal of perilymph through the round window membrane, a fibrinous precipitate formed in the endolymphatic space He interpreted this to mean a sudden increase in production of endolymph, as there was marked engorgement of the vessels which lead to the stria vascularis and occasional blood cells in the precipitate. On the other hand Perlman and Kimura (1957) in their study of experimental anoxia of the inner ear found that the stria vascularis was particularly susceptible and disintegration of the epithelial layer followed soon after production of the lesion. They found also dilatation of the strial capillaries. There seems no doubt that in the experiments of Perlman and Kimura the stria had ceased functioning and if, in Waltner's experiments, a reflex vascular disturbance resulted from the injury to the round window, then the picture of capillary engorgement, fibrinous exudate and occasional hæmorrhage would be in agreement with the findings of Perlman and Kimura, i.e. a cessation and not an increase in activity of the stria vascularis. The absence of debris in the vicinity of the normal stria vascularis may indicate not so much its power as a secretory membrane as its ability to absorb and phagocytose larger molecular aggregates.

A test of the validity of this hypothesis of the physiology and biochemistry of the inner-ear fluids would be to arrive, on the basis of its postulates, at a satisfactory explanation of certain pathological changes. In Ménière's disease the principal pathological finding is dilatation of the scala media. Other points are that the cells of the organ of Corti degenerate while the stria vascularis survives. The problem of the dilatation of the membranous labyrinth has to be reconsidered in the light of recent work. Engstrom and Liden (1957) have described some observations and experiments which show that the tension and shape of the membranous labyrinth are to a great extent dependent on the support and pressure level of the perilymph. They would consider the perilymph and endolymph as two separate hydrodynamic systems separated by a thin resilient wall. This could well be true of the semicircular canals (and perhaps of the utricle and saccule) but cannot be wholly true across Reissner's membrane. Their experiments do point to the possibility, taken together with the hypothesis under discussion, that the dilatation of the membranous labyrinth in Ménière's disease could be due to a failure of perilymphatic pressure, while the active exchange of potassium for sodium in the scala media would mean the continuance of a normal, not an increased, osmotic tension in the endolymph. Thus the only pathological lesion required on the basis of this hypothesis would be a disturbance either of perilymph production in the vascular network of the perilymphatic space, or of perilymph absorption by routes other than towards endolymph. Production of perilymph could be interfered with by a process not amounting to fibrosis but akin to the thickening of the fibrillar material of the basement membrane seen in early glomerulonephritis, or by a one-sided lesion of the nervous system supplying the vessels of the perilymphatic space—not a neuritis, but an imbalance of innervation leading to constriction of the arterioles.

Drainage of perilymph towards the spiral ligament in the outer two scalae of the cochlea and by the perilymphatic space of the vestibule and thus direct transudation to the venous system would seem to be highly probable (Altmann and Waltner, 1950, Kley, 1951) If this description of absorption of perilymph along the whole width and length of the spiral ligament is correct, then the relative pressures of endolymph and perilymph in the scalae will be at least partly dependent on the balance of absorption This may seem speculative but it should be noted that the standard description of transmission of sound stimuli involves the conversion of vibrations in the oval window to fluid movements in the incompressible perilymph This concept either entails a static fluid unchanging in volume, and probably in composition, with no inlet or outlet, or, since this is unlikely in a biological organism and improbable in any case for perilymph, it demands a dynamic flow with a system for balancing secretion and absorption Equilibration of perilymph pressure with that of the cerebrospinal fluid is conceivable in animals with a wide cochlear aqueduct, but in the human subject this explanation could not be sustained An imbalance of absorption, such as an increase of leak away from the outer scalae without a compensating increased rate of production of perilymph would lead to a relative, not absolute, increase in pressure in the scala media The continuance of this trend could produce the symptomatology of Ménière's disease in that a drop in perilymphatic pressure would lead first to an apparent increase in volume of endolymph and later to a true increase The apparent increase would occur on the basis of the model described by Engstrom and Liden, the later true increase would be due to the gradual accumulation of potassium, gradual because the rate of flow through Reissner's membrane would be diminished in proportion to the increased drain away in other directions, and accumulation because the stria vascularis in Meniere's disease remains unimpaired and, to begin with, Reissner's membrane would also be functionally intact The pressure in the endolymph need not be altered from normal Here it is worth repeating that the organ of Corti can surely not be nourished from the stria vascularis since in Meniere's disease the stria remains intact while the organ of Corti degenerates On the other hand, if the organ of Corti depends for its oxygen on perilymph, when the rate of flow through Reissner's membrane is diminishing due to reduced production of perilymph or to greater loss by other routes, then degeneration of the organ of Corti could be expected to result

Another feature of Meniere's disease could be explained on this basis As Reissner's membrane becomes more and more distended, leakage probably in sudden spurts, of endolymph might be expected to occur This would have two results, firstly sharp, probably temporary, changes in labyrinthine pressures with ensuing vestibular stimulation, and secondly, acute changes in the ionic composition of endolymph, raising sodium concentration and lowering the potassium, until the stria vascularis restored the situation. In so far as the ionic composition has a bearing on the structure and function of the tectorial membrane, sudden changes in ion concentration must be taken into account as sources of pathological irritation. If, as has been suggested (Citron, Exley and Hallpike, 1956; Davis, 1957), the fluid between the basilar membrane and the tectorial membrane is perilymph, then a sudden change in ionic composition of endolymph affecting the tectorial membrane would affect also the structures in Corti's organ at least by change in the cochlear microphonic, and perhaps by actual ion movement through the disturbed tectorial membrane and reticular lamina.

If this hypothesis of the physiology and biochemistry of the inner ear fluids has stood the test of permitting a correlated description of the pathology and symptomatology of Ménière's disease with the economy of one postulated lesion, these concepts could be put to the further tests of confirmation by animal experiment. The animal chosen should have a minimum of communication between perilymph and cerebrospinal fluid by means of an open cochlear aqueduct. Where a free communication between perilymph and cerebrospinal fluid exists there would probably be rapid equilibration of pressures and also an actual flow of fluid, and it might prove impossible to produce a sufficient imbalance of flow and pressures in the cochlea.

Another piece of information which would help to confirm this theory would be the finding, in a more chronic state of Ménière's disease, of a raised sodium and lowered potassium concentration in the endolymph.

It is perhaps worth pointing out that this hypothesis has the merit of permitting a reconsideration of the value of treatment of Ménière's disease with vasodilating agents, e.g. nicotinic acid or histamine, on a rational basis, in that the action of the vasodilator may be on the vessels serving the perilymphatic transudatory membranes. Also the question of dietary ion balance and the part possibly played by aldosterone could be-reassessed against a background of a more closely detailed physiology. It is highly improbable, on our present knowledge, that any changes brought about by a low salt diet, as recommended e.g. in *B.M.J.* (September 28, 1957) 754, have anything to do with a genuine depletion of extracellular fluid, as a means of reducing endolymphatic œdema or hydrops. It is possible, however, to suggest a means whereby dietary ion balance might exert some influence. Making reference again to the work of Johnson *et al.* (1957) mentioned earlier, it can be seen that aldosterone excretion (and secretion) is increased both with sodium depletion and potassium loading. These effects have been described also by Luetscher and his associates (1954, 1955). The dietary regimes advocated for Ménière's disease have been precisely those of sodium restriction and potassium loading. The first effect of increased aldosterone production would be to nullify the influence of the dietary changes on total body of electrolytes, but if, as seems reasonable to suggest, aldosterone has an effect on the stria vascularis similar to that which it exerts on the renal tubule—since their cellular structure is similar and their function as regards potassium movement must be presumed similar on any hypothesis—*then the effect of the dietary regimes, acting through aldosterone,* would be to facilitate sodium reabsorption and potassium secretion by the stria vascularis. In this way dietary electrolyte treatment, at a suitably early stage of the disease, could ameliorate the symptoms of Ménière's disease by aiding in maintaining a normal ionic structure in the endolymph. The more rational procedure, if this is true, is to prescribe potassium supplement rather than sodium restriction.

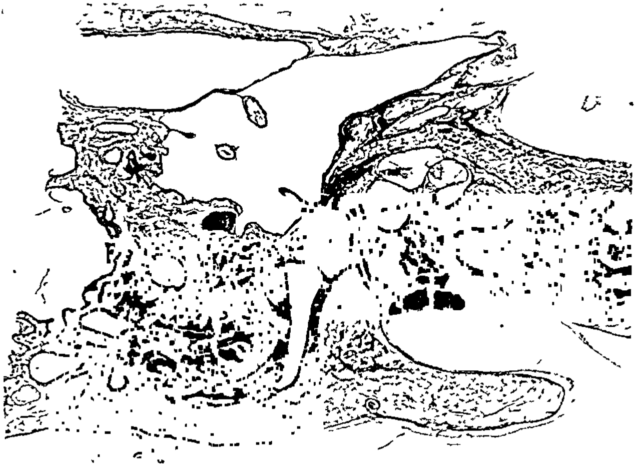

Fig 1

Low power section of temporal bone from a case of leukæmia (O 4 3 ×)

The site of origin and other factors in the physiology of the labyrinthine fluids are likely to have a considerable bearing on other pathological conditions of the labyrinth. In the cases of leukaemia of this organ described in the literature white blood cells fill the perilymph spaces, but very few are to be seen in the endolymph (Figs. 1, 2, 3 and 4 taken from

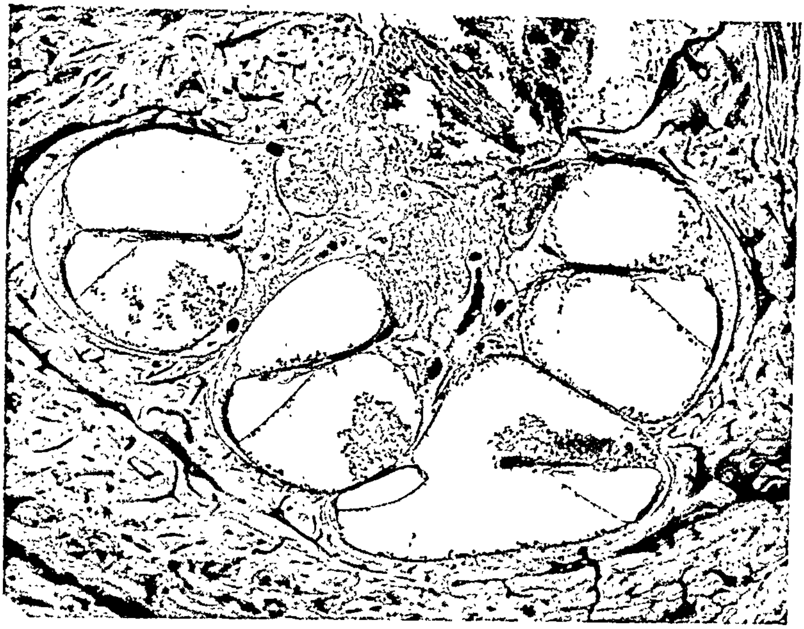

FIG. 2.
Section of cochlea (O. 10·9 ×).

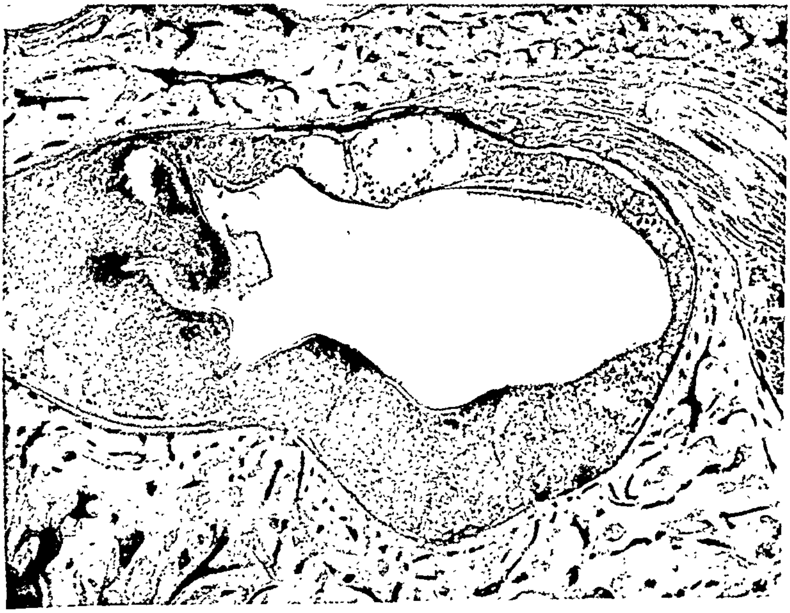

FIG. 3.
Ampulla of semicircular canal and utricular macula.

a case described by Hallpike and Harrison, 1950). If endolymph were derived from the stria vascularis the endolymph space would probably be filled with the cells escaping from the vessels supplying the spiral ligament. If, on the other hand, perilymph is a transudate the cells would be held up almost completely by Reissner's membrane and the few passing this barrier would be rapidly absorbed by the macrophages of the stria vascularis. The changes found in the labyrinth as the result of rubella are likely to be toxic in origin and here changes occur in most of the elements of the scala media

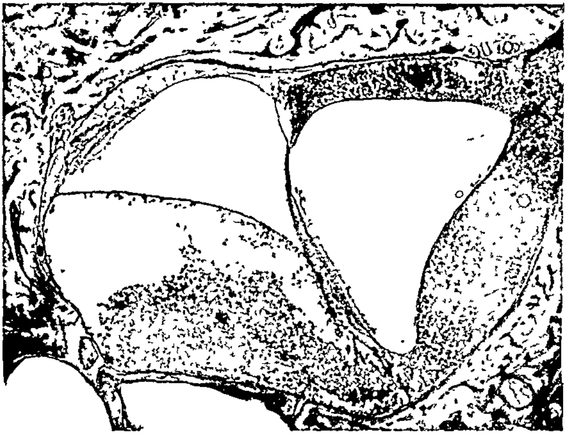

FIG 4
Stapes and macula of utricle and saccule (O 12 4 ×)

Conclusion

An active circulation of the labyrinthine fluids has been described. An attempt has been made to integrate the various factors of ionic composition and their concentration gradients with the primary requirement of any circulatory theory, viz. oxygen flow. Although some aspects are not fully accounted for, the theory is presented because of the belief of the authors that this approach gives a reasonable explanation of certain features of the histopathology of the labyrinth which hitherto have not been readily related to one another.

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NEUROGENIC TUMOURS OF THE HEAD AND NECK

REPORT OF FOUR CASES

By H MARKOWICZ and E SHANON (Tel Aviv)*

THE neurogenic tumours present a complex, pleomorphic group Many aspects of their pathology are disputed and there is no uniformity of opinion as to their origin, the distinct criteria of malignancy and the possibility of malignant transformation (Boyd (1935), Conley (1955), and Lichtenstein (1949))

Stout's (1949) classification of neurogenous tumours as modified by Conley (1955), seems to be the most acceptable one Four main groups of neoplasms occur

(1) Those originating from neurilemma benign and malignant schwannomas
(2) Those originating from nerve and neurilemma neurofibroma, multiple neurofibromatosis and malignant schwannoma
(3) Those originating from ganglionic cells benign ganglioneuroma and malignant sympathicoblastoma
(4) Those originating from paraganglionic cells paraganglioneuroma and malignant paraganglioma

The cases to be described belong to the first two groups

The majority of these tumours originate from the cell of Schwann, which is characterized by its oval nucleus and is situated on the inner surface of the sheath of Schwann, an ectodermal structure to be distinguished from the connective tissue of Henle, Maximow and Bloom (1937)

The clinical course of benign schwannomas is slow, these tumours are often asymptomatic Erhlich and Martin (1943) On the other hand, they may exert pressure upon the nerve or on the surrounding structures Microscopically, a white, firm tumour is usually found, intimately related to the nerve sheath, but not destroying nerve fibres The microscopic picture is that of slender fibres with elongated nuclei, presenting a palisade-like formation

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The malignant schwannomas, commonly referred to as neurogenic sarcoma, tend to destroy the nerve fibres and to infiltrate the surrounding tissues. Distant metastases may occur, especially in the lung. Lichtenstein (1949). Microscopically one finds whorls of slender cells; atypical œdematous cells and mitoses may be observed. At times difficulty may arise in assessing malignancy on grounds of the histological picture alone.

In the head and neck benign schwannomas have been described in most of the cranial nerves, the acoustic nerve being most commonly affected; they have been found in the tongue, palate, pharynx, larynx, in the cervical and brachial plexus, as well as in the phrenic nerve.

In the present communication we report four cases of neurogenic tumours of the head and neck, all of them of uncommon localization.

Case Reports

CASE I.—G.M., a 15-year-old girl, was born in Turkey and has lived in Israel for 7 years. Three months prior to admission (February, 1956) she noticed a non-tender nodular mass in the right side of the neck. On examination a tumour of the size of a walnut was found in the lower part of the right posterior cervical triangle. It was of hard consistency, not tender to pressure, and of limited mobility. On operation a whitish, oval mass was found on the third branch of the cervical plexus, removal of which revealed a deep depression in the nerve trunk. Pathological examination showed benign schwannoma. Careful neurological examination failed to reveal any sensory disturbance in the area supplied by the affected nerve.

CASE II.—S.H., a male patient, aged 41, was born in Poland, and for 8 years has lived in Israel. Eight years prior to admission (July, 1956) a small nodule appeared in the floor of the mouth on the left side. In the course of the years it gradually grew in size. There was no pain, but the mass interfered somewhat with speaking and moving the tongue.

Physical examination revealed (Figs. 1 and 2) a mass the size of a goose egg, to be seen both in the submandibular region and in the floor of mouth on the left side. It was smooth, hard, mobile and not tender to pressure. The clinical impression was mixed tumour of the submaxillary salivary gland. Sialography showed the gland and its duct markedly displaced. On surgical exploration of the region the submaxillary salivary gland was found to be small and atrophic, pushed aside by a large tumour. The gland and the tumour were easily removed after incising the mylohyoid muscle; the tumour seemed to be loosely attached to the lingual nerve. The pathologist's diagnosis was benign schwannoma. The operative wound healed promptly and sensation of buccal mucosa and tongue remained normal.

CASE III.—K.E., a 33-year-old male, born in Czechoslovakia, had lived in Israel for 9 years. Fourteen years prior to admission (September, 1948) the patient noticed a swelling in the right half of the tongue. He was operated upon two years later, in Czechoslovakia. The patient was told that tuberculosis of the tongue was suspected. Since then an enlargement of the tongue persisted on the right side.

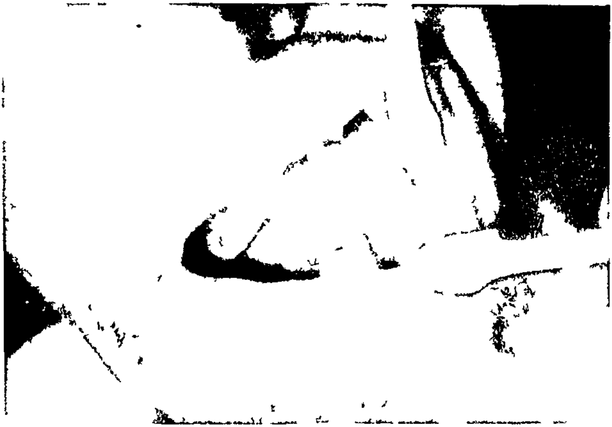
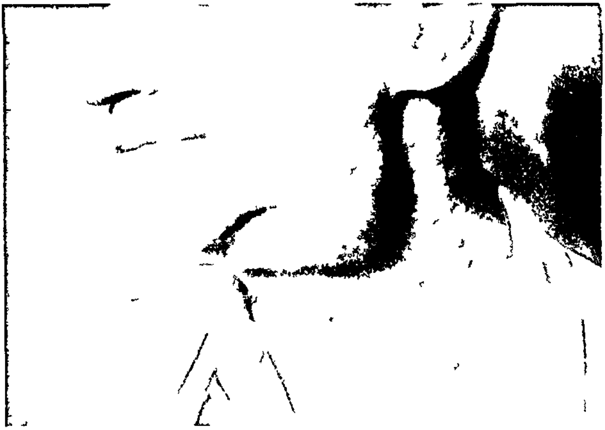


FIG 1

Benign schwannoma in the floor of mouth (Case II)



FIG

Benign schwannoma in the submandibular region (Case II)

During the last two months a large mass had developed involving the tongue, the floor of the mouth and the neck on the right side. At the same time the patient experienced difficulty in swallowing, speaking and breathing. There was hardly enough space for the tongue in the oral cavity.

On examination (Figs. 3 and 4) a soft, nodular mass of elastic consistency was found in the tongue and in the floor of mouth on the right side, bulging into the submandibular area. The tentative diagnosis was lymphangioma.

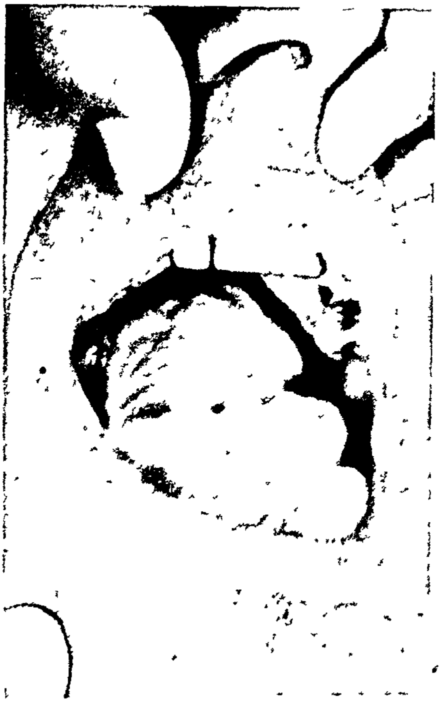


FIG. 3

Malignant schwannoma of the tongue and the floor of the mouth (Case III).

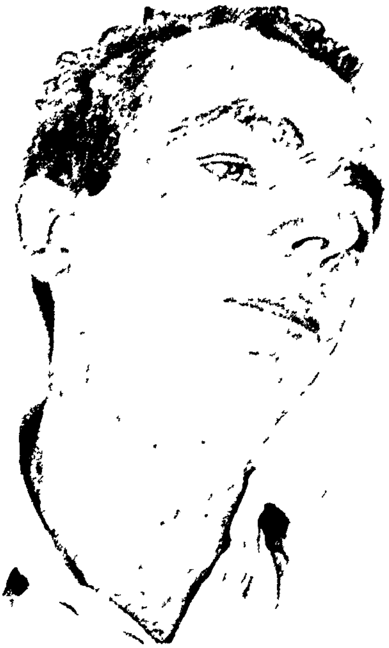


FIG 4

Malignant schwannoma in the submandibular region (Case III)

The region was explored through a submandibular incision, under local anæsthesia. An intricate network of white cords was found, diffusely infiltrating the whole region. Attempts at total removal were futile and caused excruciating pain. Therefore, only a specimen was taken: biopsy revealed malignant schwannoma.

Deep X-ray therapy was given (6000 r., air dose, in two fields). The tumour proved to be radio-resistant. The patient's condition deteriorated rapidly and difficulty in swallowing and breathing increased. The patient expired eight months after the onset of the rapid growth of the tumour.

CASE IV.—S.M., a 30-year-old male, was born in Poland, and has lived for 7 years in Israel. The family and past history were non-contributory.

The present illness started in October, 1955, with the appearance of spasmodic contractions of the eyelids, the cheek and the angle of the mouth on the right side, lasting for a few seconds and recurring about once a week In March, 1956, it was observed that his smile was asymmetrical The facial asymmetry increased gradually and the patient sought medical advice, but was reassured that his condition was a trivial and transient one In May, 1956, an E N T specialist observed signs of inflammation of the right middle ear Paracentesis revealed scanty bloody discharge

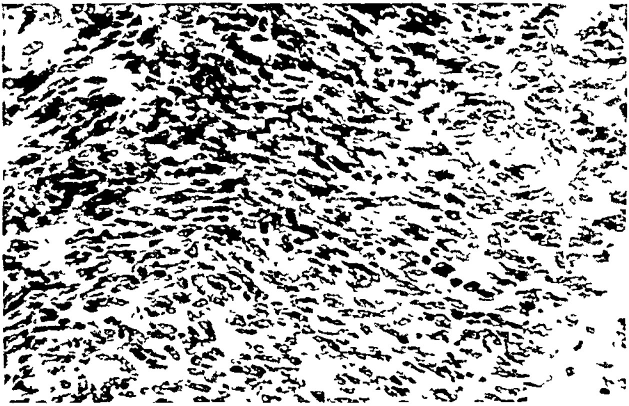


FIG 5

(4 × 47 5) malignant schwannoma of the facial nerve (Case IV) the tumour is rich in cells some of them atypical with mitoses

On admission, in June, 1956, an almost complete peripheral facial palsy was found on the right side Otoscopic examination showed several hæmorrhagic bullae on the drum puncture of one of them yielded a drop of blood A mild conductive deafness of the right ear was found The caloric reaction was normal Routine blood and urine examinations, as well as X ray examination of the chest and the mastoids showed no abnormal findings The patient was given achromycin and vitamin B_{12}, but no improvement followed In July, 1956, there was a purulent discharge from the diseased ear and in the depths of external auditory meatus polypoid tissue could be seen A biopsy specimen was taken and the pathologist suspected a tumour Exploration was decided upon, with the intention to decompress the nerve or to place a graft in case of destruction of the nerve through pressure or inflammation

In August, 1956, a radical mastoidectomy was done through a long retro auricular incision The mastoid was very cellular, the cells appearing normal

On opening the antrum, reddish tissue could be seen bulging out. Following the removal of the mastoid tip, the facial nerve, swollen and pink, was laid bare at its exit through the stylomastoid foramen. From here on the nerve was followed centrally. Between the first and the second knee the bony wall of the Fallopian canal was found to be missing, the nerve itself being replaced by a soft, reddish tumour mass. Tumour tissue filled the antrum, the aditus and the middle ear. No signs of invasion into the bony capsule of the labyrinth could be discerned. The tegmen area was intact. The tumour was removed as radically as possible, including a few millimetres of the peripheral portion of the nerve. We did not follow the nerve into the internal auditory meatus. Nerve grafting was deemed not feasible because of the localization of the defect and the doubtful radicality of our procedure. The operative cavity was left widely open.

The pathologist found the tumour to be rich in large spindle cells, arranged in a disordered fashion. Few mitoses and atypical cells were seen. The diagnosis was malignant schwannoma (Fig. 5). Three weeks after operation a tarsorrhaphy was done on the right side. The mastoid cavity was lined with a Thiersch graft leaving the affected region free for future inspection and control. Fifteen months after the operation no signs of recurrence could be seen.

Comment

Four cases of neurogenic tumours in the head and neck were described, two benign schwannomas (one in the floor of mouth and the other in the cervical plexus) and two malignant ones.

The first of the malignant tumours developed in the tongue and in the floor of mouth. According to the history it may be an example of malignant transformation in a benign tumour. The second one affected the facial nerve. This appears to be the third reported case of malignancy in the intratemporal portion of the VIIth nerve, and the first one reported affecting its horizontal segment. Perusal of the literature (Kettel (1950), Guttman and Simon (1951), Bogdasarian (1944), Lundgren (1947), and Loeliger (1947)) as well as the history of this case show that one should exercise caution in diagnosing Bell's palsy. When dealing with peripheral facial palsy developing insidiously over a period of weeks or months and unaccompanied by pain, one should consider the possibility of a facial nerve tumour. In such cases an early exploration might reveal the true nature of the palsy. A benign tumour may be excised cautiously. In case the decompression proved ineffective a nerve graft should be inserted at a later date. When a malignant tumour is diagnosed, we think that one should be satisfied with a relatively conservative operation, as the grade of malignancy of these tumours seems to be low; sacrificing important structures such as the labyrinth would hardly be justified. Nerve grafting should be done whenever possible. It appears of advantage to leave the retroauricular cavity widely open for periodic examination. Radiotherapy is of no avail in these tumours.

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SOME PROBLEMS OF TYMPANOPLASTY*

By P. H. BEALES (Doncaster)

THIS paper is based on the experience I have gained over the last two years from the performance of 142 plastic operations on the middle ear in patients suffering from chronic suppurative otitis media.

I believe these new techniques are a remarkable advance in the surgical treatment of a common and disabling disease and replace in most cases the existing methods of surgical treatment which in any case have never been wholly satisfactory for the patient or the surgeon; the modified radical operation is often inadequate and does not always remove the disease in the middle ear and the classical radical operation is a destructive operation which is today rarely justifiable.

I wish to discuss the problems I have encountered under four main headings:

Problems of Tympanoplasty

1. Problems of selection of cases.
2. Problems of technique, including two-stage operations.
3. Problems of after care and the behaviour of skin grafts.
4. Problems of late complications, including acute otitis media after tympanoplasty.

1. *Problem of selection of cases for Tympanoplasty.*

About 75 per cent. of my cases have been treated by tympanoplasty and 25 per cent. by myringoplasty.

I have divided my cases for operation into two groups:

1. Children and young adults up to age 35 years.
2. Middle aged patients.

In group 1 there is in nearly all cases adequate cochlear function and I believe that if this is so whatever degree of sepsis is present and however advanced it is if an *adequate and correct operation* is performed one can achieve a dry and functionally useful ear. In group 2, the older age group, selection is more difficult as many of these patients have a nerve deafness and in a proportion the grafts become sclerotic and rigid and the functional improvement is less certain. If one does fail to get a marked functional improvement in this type of patient one does at least achieve a dry ear in the majority as the eustachian tube is sealed off from the mastoid cavity by the skin graft.

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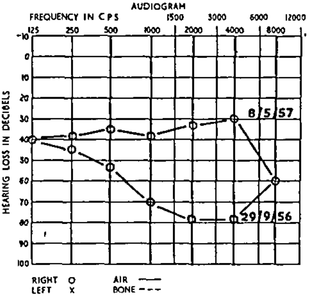
* Paper read at Section of Otology of the Royal Society of Medicine on June 27th, 1957, at the Summer Meeting at Bristol

I believe that if one decides that a tympanoplasty is not indicated but that the mastoid requires full exploration one should in all cases graft the tympanic cavity with a full thickness skin graft.

Finally, we all meet from time to time the unfortunate patient who has had bilateral radical operations in the past, has a severe hearing loss and asks if something can be done to improve the hearing. I have had some experience of this type of case and one can get good functional results in a small proportion of them, but I think that it is impossible without exploration of the middle ear under magnification to make a prognosis as to functional improvement from tympanoplasty performed after a radical operation. If there is gross scarring of the window regions an improvement of function is unlikely to take place although it is possible to use mucosal grafts after removal of the scar tissue.



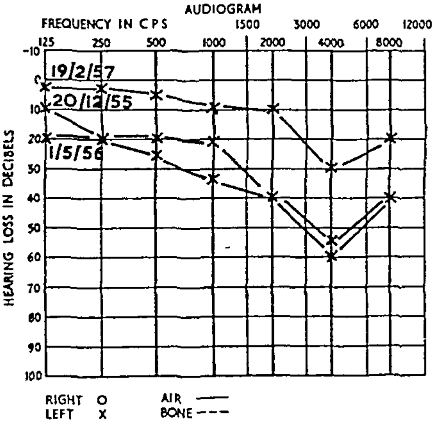
Revision, right radical mastoidectomy October 1956

Choosing the correct operation

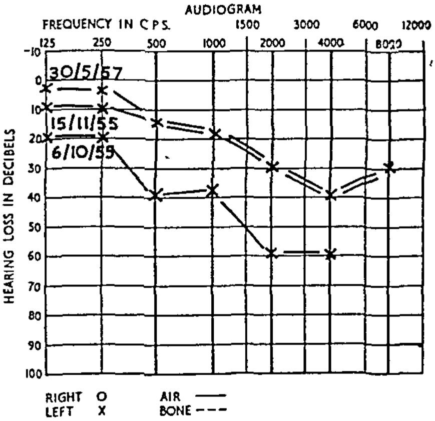
We have at our disposal four common operations and two that are rarely required.

Type 1 and Type 2 operations

These operations are applicable to cases of middle-ear disease with an intact and functioning chain and minimal middle-ear or mastoid disease. This type of operation consists essentially in an attico-antrostomy reflection of the drum forwards, examination of the ossicular chain and testing of its function and removal of disease with preservation of the sound-conducting apparatus. The defect in the drum is then repaired with a full thickness graft extending over the bridge which is left if possible to prevent the graft from adhering to the incus. If it is necessary to remove the bridge one can get a good functional result provided that the incus is left with its covering of mucosa. These operations give good results in selected cases with minimal disease and the type 1 operation can be performed in all cases where a myringoplasty is indicated and is probably a better operative procedure for the cautious. There is always an element of doubt in carrying out a blind procedure such as a myringoplasty although I think it is a good operation for the right case.



Left Tympanoplasty, Type 1, 12.12.55 Audiograms show progressive hearing gain.



Type 1. Tympanoplasty, 7.10.55

Type 3 and Type 4 Tympanoplasty

These types of operation are performed in cases of gross mastoid disease and when the ossicular chain is interrupted and in my opinion they are the most important of all the plastic operations on the middle ear for the following reasons:

A. The operations are truly "radical" in the sense that a most meticulous operative toilet with removal of all disease is first carried out under magnification in all cases. If cholesteatoma is present one can be quite certain that it has been removed.

B. Adequate toilet of the window regions is possible.

C. The tympanic reconstruction is not performed until one has an ear that is fit for it. In some cases it is not performed until 14 days later.

D. The eustachian orifice is closed off from the mastoid cavity by the graft which is able to heal properly and produce a dry ear.

E. Finally, remarkably satisfactory functional improvement can be obtained from these operations.

Two-stage operations.

In cases of gross sepsis and in particular when it has been necessary to remove a good deal of middle-ear mucosa in the original toilet, a delayed reconstruction during which time the patient receives antibiotic treatment is of value. Middle-ear epithelium regenerates rapidly and by the time the graft is put into position the tympanic cavity is found to be covered with what appears to be a normal epithelium even if it had been largely denuded of its mucosal covering at the first operation. I have been impressed at the satisfactory results I have obtained from two-stage operations and perhaps one should resist the temptation to do a one-stage operation and do more of them.

In concluding this part I will mention the basic principles for the successful take of a full thickness skin graft in the tympanic cavity. They are seven:

1. Thin graft, no subcutaneous fat.
2. Adequate contact of graft with vascular bed until it ceases to be a parasite.
3. Absence of blood beneath graft.
4. Absence of sepsis.
5. Adequate firm pressure until vascularized.
6. Absence of trauma to graft when cut at operation.
7. Absence of bone chips beneath graft.

We are able to control all these factors but the omission of one or more of them will lead to partial or complete failure of the graft to take. If this occurs, except as a rarity, in a site such as the tympanic cavity which is an extremely favourable one to graft, it can only be interpreted as the result of faulty technique on the part of the operator.

Before leaving the question of technique I will mention some operative mistakes which occurred in my cases. On two occasions I have dislodged and removed the stapes with the sucker. This did not have a disastrous effect on hearing as the oval window was immediately closed with the skin graft.

On one occasion I opened the membranous canal with the drill when opening the attic and on one occasion the round window membrane was lifted and blood entered the labyrinth. This unfortunately produced a severe labyrinthitis and the patient's hearing fell off badly.

Problems of after-care

In my early series of cases I grafted the mastoid cavity as well as the tympanic cavity. Analysis of these cases showed a prolonged healing time in many of them.

The cause of this in nearly every case was due to delayed healing of the mastoid cavity and in the remainder with delayed healing it was due to inadequate technique with failure to remove all disease at the original operation.

If one examines sections of the skin grafts removed from a patient subjected to tympanoplasty some time previously it becomes clear why this occurs. (Figs. 1, 2.)

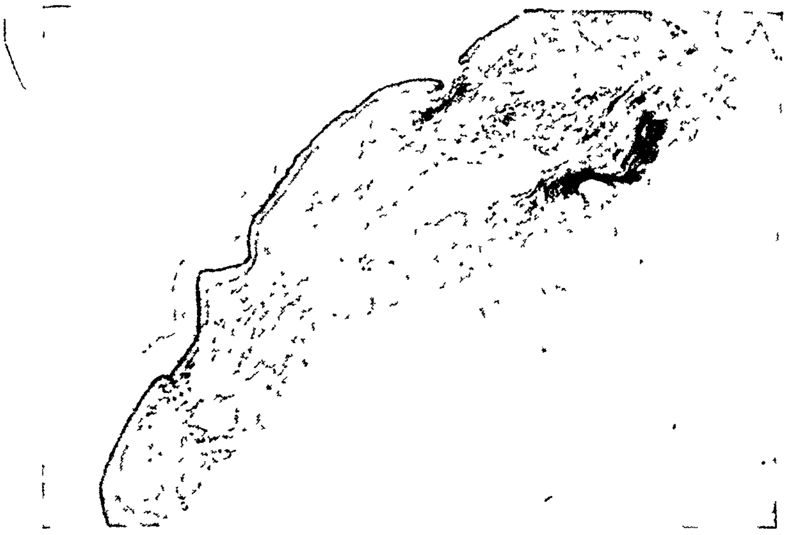


FIG 1

SPECIMEN I SKIN FROM MASTOID CAVITY

This specimen consists of slightly keratinizing stratified epithelium and an underlying zone of connective tissues No dermal papillae or appendages are present and the biopsy material is indistinguishable from any mucosal membrane covered by stratified epithelium There is a fairly dense infiltrate of lymphocytes and histiocytes in the zone bordering the epithelium, which also contains some amorphous calcareous debris Practically no elastic fibres are demonstrable in the tissue.

It is evident that skin applied to the tympanic cavity survives as true skin retaining all the specialized structures, such as hair follicles and sweat glands, while skin applied to the mastoid cavity atrophies and part of it sloughs away. I do not know the reason for this. It is not due to the graft being applied over air cells as in the majority of cases the mastoids are sclerotic and it is possible to remove all epithelium, thus producing a surface which should take a graft well.

I now apply the graft to the tympanic cavity only and leave the mastoid cavity to heal aided by local skin flaps. Healing is often more rapid with this technique.

Inflation of the middle ear after tympanoplasty

As the graft for a considerable period after it is applied to the tympanic cavity must have a precarious blood supply I have hesitated to inflate the middle ear by politzerization or catheterization for a period of some weeks after operation. If one uses gelatin foam there is little tendency for the graft to adhere to the promontory and owing to the shape of the tympanic cavity there is a natural tendency for a cavity to form beneath the graft. I believe that routine inflation of the middle ear soon after operation damages the graft and produces eventual scarring with a sclerotic graft. I have very few cases with the production of sclerotic grafts, but believe I would have many more if I practised routine inflation and premature stretching of the graft.

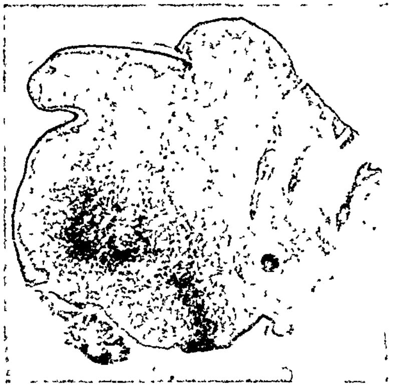


FIG 2

SPECIMEN 2 SKIN FROM EDGE OF PERFORATION

Skin wi[illegible]
At the [illegible]
ciliated [illegible]
a single layer of cells resembling the epidermal basal layer Elastic fibres are present in the parts of the specimen lined by a stratified squamous epithelium, but absent from the layer covered by columnar epithelium

Lining of the skin graft

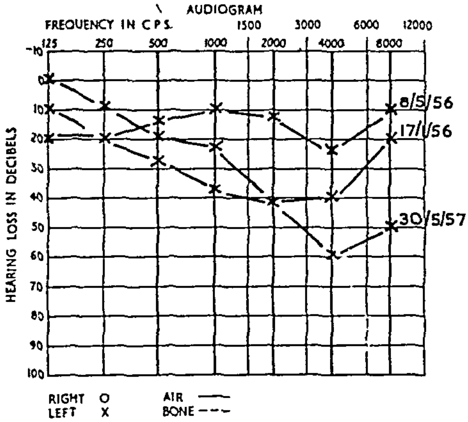
The plastic surgeon always likes to line the raw surface of skin grafts to prevent contraction. I do not think this principle applies to skin grafts in the tympanic cavity as there is a natural tendency for the middle-ear epithelium to grow rapidly over the raw surface on the inside of the graft and one will get a much thinner graft if this is allowed to happen naturally.

P. H. Beales

Problems of Late Complications after Tympanoplasty

Later perforation of the graft

Small pin-point perforations of the graft occur quite frequently, in about 15 per cent. of cases. They usually appear within a few weeks of the operation and are more common in older patients than younger ones. They indicate that the blood supply of the graft is still poor and they should be left strictly alone and no attempt made to stimulate proliferation of epithelium by touching up the edges, etc., or this will result in a larger perforation and possibly loss of the skin graft. I have had little experience of large perforations of the graft and if they occur frequently they indicate inadequate technique. It is no exaggeration for me to say that total loss of the graft is extremely rare. The great majority have a 100 per cent. initial take.



Left myringoplasty 6.4.56 Audiograms show high tone loss.

Recurrent otitis media

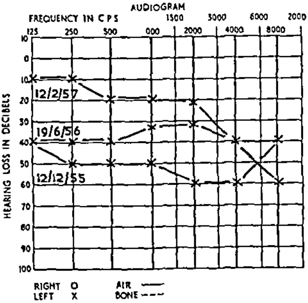
I have seen this occur in about four cases. All were treated with antibiotics and three resolved completely. The remaining case developed a labyrinthitis and required re-opening. A type 1 operation had been performed and at the second operation mucopus was found in the middle ear. No late complications of this sort have occurred in any of the total tympanoplasties. This is of great interest as many of these cases had very septic ears at the original operation.

Otitis media after tympanoplasty presents a different picture to otitis media as we understand it. There seems to be no tendency for the graft to perforate and pain is less severe.

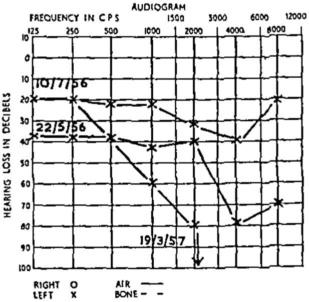
Late fall-off in hearing after tympanoplasty

A number of cases treated by all techniques, myringoplasty, partial and total tympanoplasty, have shown an initial hearing improvement to be followed by a gradual fall-off in the higher tones This process may take a year or more to become apparent

I do not know the exact reasons for this but one must take into account possible failure of the eustachian tube, late sclerosis of the graft and low grade infection in the middle ear causing cochlear damage



Left Type 3 Tympanoplasty 8 1 56
Audiograms show slight high tone loss



Type 3 Left Tympanoplasty 25 5 56
Audiograms show severe high tone loss

In conclusion, my experiences over the past two years have convinced me that these new techniques offer a remarkable advance in mastoid surgery and replace the older techniques The only criticism of them is that they are not safe for the patient I have no evidence that they are not safe from my experience up to now but only the fullness of time will show if this is true

I do not believe that one should carefully select a promising case for tympanoplasty but that one should adapt the different techniques to the pathology found at the primary operation No one should embark upon a minor operation such as myringoplasty unless they are prepared to change the operation if necessary and do a total tympanoplasty as one can never be certain of the exact pathology until the middle ear has been examined under magnification

I think that the partial operations including myringoplasty are the most difficult operations to select and although I believe they have a part to play in the surgery of chronic suppurative otitis media it may be proved in the future that total tympanoplasty is the safer and more lasting operative procedure

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CLINICAL RECORDS

A CASE OF FAMILIAL HÆMORRHAGIC TELANGIECTASIA ASSOCIATED WITH PULMONARY ARTERIOVENOUS FISTULA

By D F N HARRISON (London)

It is well known that familial hæmorrhagic telangiectases may be associated with isolated or multiple arteriovenous fistulae in the lungs However the condition is rare and the following case would thus appear worthy of reporting

Case Report

Charles B , aged 41, was first seen in the Out-Patient Department of Guy s Hospital on *June 19th, 1956*, complaining of severe epistaxis since childhood The bleeding had gradually increased in severity and his nose had been cauterized on several occasions His mother suffered from a similar condition and is included in a series previously reported (Case IV—Harrison)

Examination revealed widespread telangiectases and he was advised to come into hospital for investigation prior to commencing œstrogen therapy this he refused to do!

His next attendance at the Out Patient Department was on *July 3rd, 1957* when he said that the bleeding had become much worse In addition he had noticed some dyspnœa over the past year, this had also increased in severity until he was only able to walk about twenty yards without resting However he was able to lie flat in bed and had no palpitations cough, hæmoptysis or chest pain He was admitted to the medical wards of the hospital under Dr R Kauntze for investigation

Examination

A rather anxious lightly built man with some marked peripheral cyanosis but no clubbing

The heart was not enlarged but the sounds were not easily heard because of widespread emphysema The blood pressure was normal and the pulse regular

A sharply localized systolic murmur was heard to the right of the midline in the sixth interspace some one inch from the sternum Telangiectases were present on the lips, hard palate dorsum of nose and both sides of the nasal septum, fingertips and on both pinnae

INVESTIGATIONS

As it was intended to commence treatment with ethinyl œstradiol estimations of hepatic function were carried out together with values for serum electrolytes and Hæmoglobin levels all were within normal limits

100 per cent. suggests that the diffuse pulmonary emphysema is the major factor in the unsaturation observed on room air breathing. Nevertheless this should not be interpreted as meaning that no blood flows unaerated from the pulmonary artery to the pulmonary veins through the fistula, since there are technical limitations to the determination of hæmoglobin saturation by the Haldane method on 100 per cent. oxygen breathing. The calculated saturation should be in the range 105, 108 per cent. by the Haldane method on 100 per cent. oxygen breathing.

The slightly elevated cardiac output is on the basis of a narrow arteriovenous difference, with a normal oxygen consumption. The high output state sometimes seen in pulmonary emphysema is usually on the basis of elevated oxygen consumption with a normal arteriovenous difference. This may mean that the slight elevation of cardiac output here is a circulatory adaption to the right to left shunt in the lungs.

The clinical symptomatology is undoubtedly due primarily to the diffuse pulmonary emphysema. It is unlikely that the physiological effects of the arteriovenous shunt itself are very significant clinically. However, if there is actually an absence of pulmonary circulation to a large portion of the right lung which is ventilated, as the angiogram suggests, such useless ventilation of non-perfused lung may be an important factor in the disability of a man with virtually no ventilatory reserve. It would probably be necessary to perform differential bronchospirometry in order to decide whether there is sufficient function of the left lung to allow right thoracotomy and resection of the non-functional portions of the right lung.

In view of these findings it was felt that there was no immediate indication for operative interference and the patient was discharged from hospital on a daily dosage of 0·25 mg. of ethinyl œstradiol.

Comments

The usual vascular abnormality in familial hæmorrhagic telangiectasia is a localized thinning in the capillary walls. However there may also be visible dilation of arterioles and capillaries and this can lead to the formation of arteriovenous aneurysms, especially in the lung.

In the case reported it is probable that this lesion had been present for a considerable period of time and it is only the development of pulmonary emphysema that has drawn attention to the fistulous communication existing in the pulmonary circulation. No clinical or radiological evidence could be found of any similar condition in the patient's mother.

Acknowledgments

I would like to express my thanks to Dr. R. Kauntze and to Mr. Philip Reading for permission to record this case.

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EMBRYONIC SARCOMA (RHABDOMYOSARCOMA) OF THE NASOPHARYNX PRESENTING WITH FACIAL PALSY

By C A HOLBOROW and LESLIE L R WHITE (London)*

Introduction

THE case reported below of a nasopharyngeal neoplasm in a child is of considerable interest in two respects, namely the unusual nature of the tumour and its mode of presentation

Clinical History

Maria, a girl aged four years and one month, was the youngest, until a week before admission, of a healthy and prolific Greek Cypriot family

She first came under medical attention for her present condition on *March 19th*. Two weeks previously she had had a cold During the last two days her face had appeared "peculiar" and there had been some pain in her left cheek On examination there was a left facial palsy of a lower motor neurone type, some swelling of the left cheek and injection of the left tympanic membrane (Fig 1)

A diagnosis of Bell's palsy was made

On review (*March 24th*) there was considerable swelling of the face and a discharge from the left nostril She was given penicillin until *April 4th*, when she was referred to the E N T Department for opinion on her nasal sepsis

On examination a plump, rather miserable child speaking Greek with only a few English words The left side of the nose was filled with pus, the left cheek was swollen and the left side of the soft palate was pushed down by a large, firm, painless mass which was palpable in the post-nasal space The tonsils were large but normal The tympanic membranes and hearing were normal The tonsillar glands were enlarged as were glands in the left posterior triangle of the neck

Cranial Nerves

- Left facial palsy of a lower motor neurone type
- Absence of lachrymation from the left eye
- Diminution of sensation over the left trigeminal (maxillary division) area
- Left corneal reflex absent
- Left side of soft palate paralysed

The eyes showed normal movements, reflexes and fundi

The remaining examination was normal

The child was admitted under a diagnosis of nasopharyngeal growth

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* In receipt of a grant from the British Empire Cancer Campaign

Investigations to confirm the diagnosis

X-rays of Skull (Fig. 2)

There was extensive erosion of the base of the skull, the area involved including the greater wing of the sphenoid, petrous apex and the sphenoid sinus together with the posterior ethmoid region on the left side. The area of erosion extended into the bony eustachian tube. There was an associated hemispherical soft-tissue swelling projecting into the nasopharynx from the left.

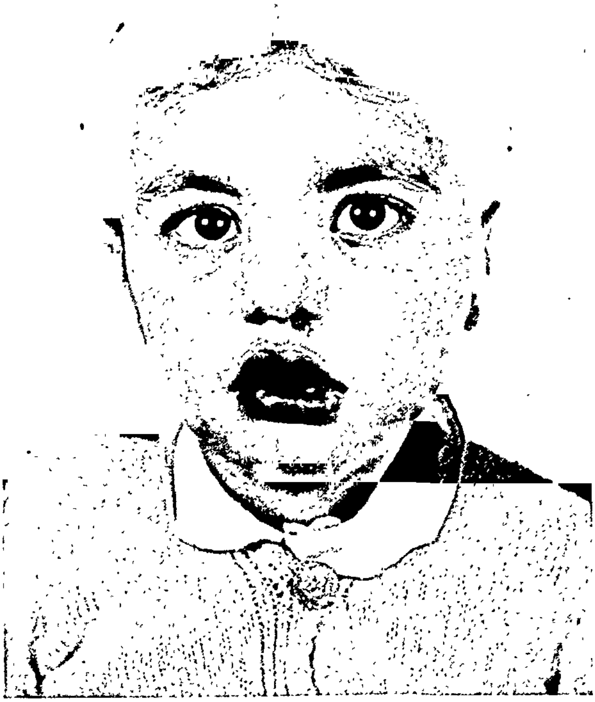

FIG. 1.

Appearance of patient, showing facial palsy and swelling of cheek.

X-ray of her chest was normal.

Blood, Hb. 91 per cent. W.B.C. 8,900.

On *April 6th* an examination under anæsthesia showed a large smooth hemispherical tumour filling the left half of the nasopharynx. A biopsy was taken.

Microscopic examination of the biopsy fragments revealed neoplastic infiltration of a highly cellular type and pleomorphic appearance. The architecture, cytology and presence of a small number of probable rhabdomyoblasts suggested the diagnosis of embryonic sarcoma (rhabdomyosarcoma) of the nasopharynx (Fig 3)

On *April 10th* radiotherapy was commenced This was continued until *May 7th*, when it was abandoned because of general deterioration, low platelet count and pyrexia At this time the palatal palsy had disappeared and the growth was much smaller There were no additional abnormal signs and X rays of the long bones showed no metastases On *May 29th* the child, though very weak, was allowed home at the request of the parents

Fig 2
X ray of base of skull

When she was readmitted on *June 5th* the swelling in the nasopharynx was not noticeably larger and the facial palsy was unaltered There was however a left proptosis, total ophthalmoplegia and blindness, with a left complete trigeminal palsy

A week later the left ear, which till then had been normal, discharged blood and pus

Death took place on *June 19th*

Autopsy Report

Externally the skull was generally of normal size and configuration, but the left zygomatic arch was slightly more prominent than the right being expanded and nodular at its posterior extremity There was some ill-defined facial swelling in the left pre auricular region the left eye was mildly proptosed and the globe disorganized, with destruction of the cornea. An adhesive fibrinous left-sided conjunctivitis was present. The remainder of the body appeared normal apart from some wasting and the presence of small subcutaneous parasternal nodules at about the level of the second costal interspaces.

Internally: the posterior wall of the nasopharynx was displaced forwards on the left side by a somewhat lobular and ill-defined mass of pale tumour tissue, partly invested by the palatal and pharyngeal musculature, and bounded laterally by the ascending ramus of the mandible The tumour extended upwards, filling the infra-temporal fossa, and had destroyed the floor of the left middle fossa, forming a dome-shaped subdural elevation approximately 3 × 4 cm. in the latter region. Medially this subdural mass had encroached on and partly destroyed the sella, so that it invested the pituitary gland; posteriorly there was extension to the petrous portion of the temporal bone, which was expanded and replaced except in its postero-lateral portion. The middle-ear cavity was largely obliterated and filled with neoplastic tissue, but the inner ear appeared intact The mastoid air cells contained yellowish material, probably inflammatory granulation tissue. The left eustachian tube could not be defined except in its cartilaginous portion, where it appeared intact and patent

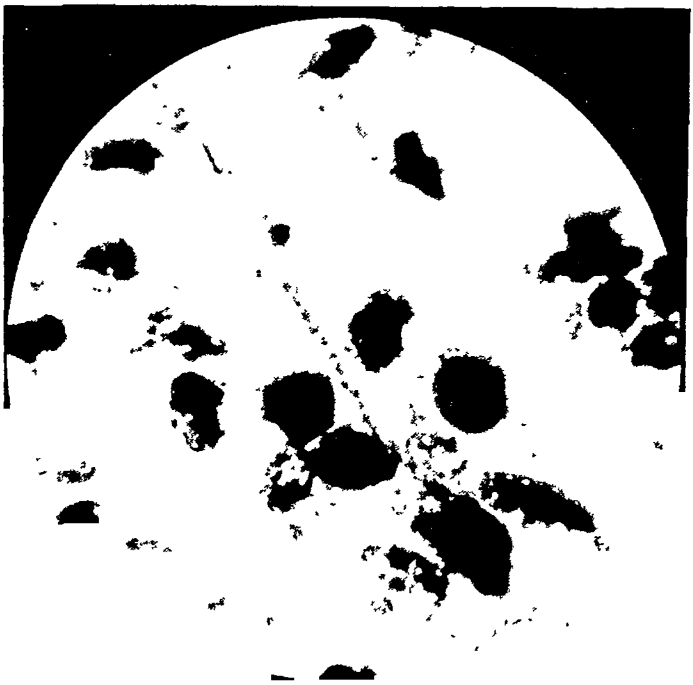

FIG 3

Photomicrograph of biopsy from primary tumour, showing a slender rhabdomyoblast with cross-striation (Mallory's phosphotungstic acid hæmatoxylin × 1600) but embedded within the primary tumour. The Vth left cranial nerve, between its point of emergence from the brain-stem and entry into the dura, was expanded around an ovoid nodule of tumour tissue 1·5 cm in maximum diameter. A similar but slightly larger nodule interrupted the course of the left VIIth and VIIIth cranial nerves immediately proximal to their entry into the internal auditory meatus (Fig. 4).

The alimentary system showed no significant abnormality apart from an ill-defined variegation of the liver, suggestive of early but diffuse metastasis.

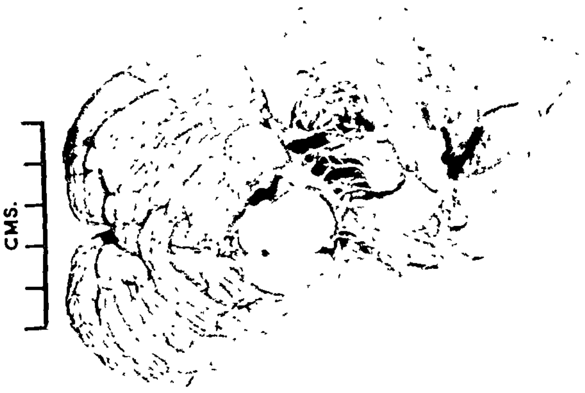

FIG 4

Autopsy specimen showing expansion of the temporal bone and tumour nodules on the left Vth and VII-VIIIth cranial nerves

Both pleural sacs contained approximately 200 ml. of fluid, clear and yellowish on the left side but blood stained on the right There was extensive irregular deposition of tumour beneath the parietal pleura (chiefly in the paravertebral regions) and on the upper surface of the diaphragm posteriorly. All lobes of the lungs were studded with numerous nodules of tumour up to 1·5 cm. in average diameter, both beneath the pleura and at deeper levels, where they occasionally surrounded a bronchus or blood vessel

The mediastinal and internal mammary lymph nodes were infiltrated by tumour tissue, as was also a solitary pre-tracheal node The thymus was atrophic. The spleen was rather more than twice the expected weight for the age and of firm consistency; on section the pulp was congested and the lymphoid follicles prominent.

In addition to the destruction of the skull produced by the primary tumour, there were a few small scattered subperiosteal metastatic plaques on the outer aspect of the calvarium. Multiple secondary deposits replaced the marrow in the sternum, vertebral bodies and femur, the tumour tissue in the former sites showing appreciable subperiosteal extension.

Other systems appeared macroscopically normal apart from the right adrenal gland, in which there was a small metastasis.

Microscopy

The primary tumour showed considerable variation in its morphology in different regions. In general it was highly cellular and considerable mitotic activity was present. The cells for the most part contained moderately large chromatic nuclei, with scanty delicate surrounding cytoplasm which formed slender processes sometimes apparently anastomosing with those of neighbouring cells. Occasional cells had more abundant and eosinophilic cytoplasm, but though these were suggestive of rhabdomyoblasts, in none was unequivocal cross-striation demonstrable In the most cellular areas little arrangement was discernible, but in regions more particularly where there was a moderate

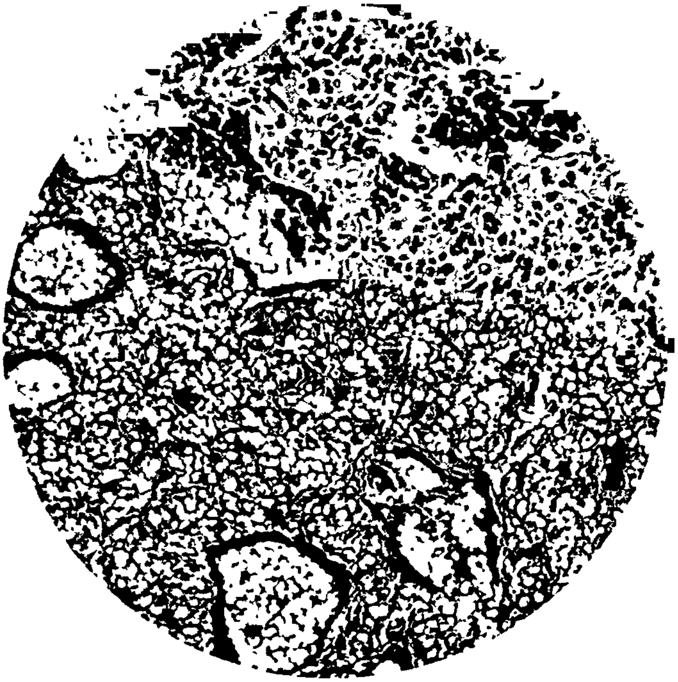

Fig. 5.

Photomicrograph of pulmonary metastasis Whilst the majority of the tumour cells (and especially those in the lymphatics) are very primitive, scattered cells have abundant eosinophilic cytoplasm (H. and E. × 200.)

amount of delicate reticulin deposited between the cells, the latter assumed a fusiform shape and showed some orientation into interlacing strands Focal necrosis was present and occasional areas were replaced by hyalinized acellular collagen, possibly as a result of radiotherapy Marginally, in the pharynx the tumour had infiltrated and engulfed surrounding tissues, including fat and muscle Section of the left eustachian tube near its pharyngeal end showed a small focus of submucous tumour tissue, but no generalized infiltration

Histological examination confirmed the presence of metastases in the sites enumerated under the gross autopsy findings They consisted in general of very primitive cells showing little or no differentiation, but in the lung (Fig 5) there were some clusters of cells with abundant eosinophilic cytoplasm which, although not displaying cross striation contained numerous glycogen granules, presumptive evidence that they were rhabdomyoblasts In this site, in addition to ill defined (presumably blood borne) metastases there was tertiary and extensive lymphatic permeation, with resulting involvement of the subpleural tissues A somewhat surprising finding was the presence of extensive colonization of the sinusoids of liver, spleen and a small spleniculus by primitive tumour cells

Discussion

(a) *The Nature of the Tumour*

The diagnosis of "embryonic sarcoma ' requires explanation The term has been suggested (White, 1952, 1955, 1957) as a designation for a type of tumour which occurs chiefly in childhood in the urogenital tract and to a lesser extent in the nasopharyngeal region and elsewhere The tumour consists basically of an immature or "embryonic' mesenchyme which exhibits a variable degree of differentiation into striated muscle, connective tissue and possibly smooth muscle Because of the resulting variations in structure, a variety of names has been applied to such tumours including myxoma, myxosarcoma fibrosarcoma and, where the striated muscle constituents have been recognized rhabdomyosarcoma The neoplasms appear to develop over a variable portion of a field of origin in submucosal regions related to epithelium of particular embryological derivations In the urogenital region, the tumours commonly exhibit initially a comparatively innocuous structure and extend only locally for a time though where survival is sufficiently long a rapid growth phase may occur, characterized by anaplastic cytological changes and metastatic extension Those tumours which we have observed in relation to the nasopharynx have however shown greater aggressiveness, early metastasis and death

In the present instance, histological diagnosis was not an easy matter However, the presence of scanty elongated cells with appreciable cross striation in the biopsy specimen (Fig 3) and the occurrence of large glycogen rich cells in some of the pulmonary metastases (Fig 5) established the nature of the tumour with reasonable certainty

In some respects, notably the extensive subpleural invasion, and the colonization of liver and spleen, the behaviour was reminiscent of reticulum-cell sarcoma, but these rather unusual features may have been the result of the extremely rapid growth rate and consequent lack of differentiation in what was after all a mesenchymal tumour

(b) *Modes of Clinical Presentation*

The various ways in which nasopharyngeal tumours may present were reviewed by Godtfredson (1947) Ophthalmo-neurological symptoms were a presenting feature in only 16 per cent of cases, and of these by far the most frequent were due to trigeminal involvement By the time of diagnosis the corresponding figure was 38 per cent, and a much wider range of cranial nerves was affected, Nevertheless, at all stages of evolution the facial nerve was amongst the least frequently implicated It is, therefore, of interest that in the small number of rhabdomyosarcomatous tumours of the nasopharyngeal region so far reported, facial palsy has been of relatively frequent occurrence This unusual feature is, however readily explicable since some of these growths appear to have arisen in the middle ear, or to have extended into the cleft, apparently by way of the eustachian tube In the present instance it is remarkable that despite the fact that such a palsy was the initial feature, there was no clinical evidence that the middle ear was implicated until the terminal stages of the illness The possibility that the facial nerve was involved peripherally (i e external to the stylo-mastoid foramen) is unlikely in view of the fact that there was absence of lachrymation on the affected side, indicating a lesion at or proximal to the geniculate ganglion, moreover, there was no definite evidence that the nerve was implicated in its extracranial course at autopsy A theoretical possibility is that of tumour extension along the Vidian and greater superficial petrosal nerves Such spread, by perineural lymphatic paths, has in fact been suggested to explain neural lesions associated with nasopharyngeal tumours (Godtfredsen, *loc cit*) However, there was no positive evidence for this hypothesis in the present instance, and such a mode of spread is not in general characteristic of rhabdomyosarcomatous tumours

Summary

1 An instance of embryonic sarcoma (rhabdomyosarcoma) of the nasopharynx in a four-year-old girl is reported

2 The mode of presentation, namely with facial palsy, was unusual

3 The nature of this type of tumour, and the symptomatology of nasopharyngeal neoplasms, are discussed briefly

Acknowledgments

We should like to record our thanks to Mr James Crooks, under whom the patient was admitted, to Dr Martin Bodian, who reported on the biopsy specimen, and to Mr Derek Martin for the illustrations

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EXTRACRANIAL NEURILEMMOMA OF THE VAGUS

By J. F. O. MITCHELL (Dundee)

BONY lesions of the petrous pyramid are uncommon, and notoriously difficult of access; they may be primary, or more frequently arise by metastasis.

Clinical features are the result of involvement of the adjacent cranial nerves or brain-stem. The following case, discovered during routine investigation of a right vocal cord paresis, exemplifies the progressive neurological involvement, and the difficulty of diagnosis and of access.

Mrs. B.G. was first seen in 1949 (age 51 years) on account of deafness progressive since schooldays; examination showed intact drumheads and a bilateral conductive deafness (Rinne negative right and left) with some right-sided perceptive loss also, attributed to a blow on the head at school. A Medresco insert hearing aid was provided.

On *2.4.55* she returned complaining of huskiness and choking turns for some six weeks, attributing these to a heavy cold. Examination then showed a complete and flaccid paralysis of the right vocal cord, normal ear-drums, and a clear nasopharynx; hearing tests revealed a now total deafness on the right, acuity on the left being limited to a conversation voice at a distance of six inches.

X-ray of: { Chest was normal.
Skull showed destruction of the inner part of the right petrous bone (Fig. 1*a* and *b*).

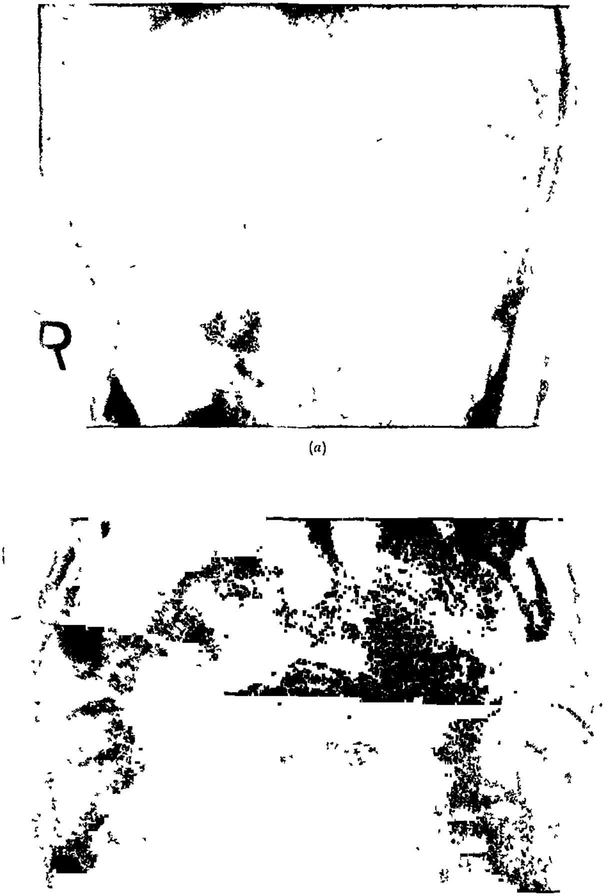
Neurological examination showed no other lesion, and ophthalmic examination was negative.

On *21.4.55* examination showed, shining through the right tympanic membrane, a pink mass in the hypotympanum, and filling the lower third of the middle ear; this was certainly not present on *2.4.55*.

By *18.7.55* this mass was, if anything, more prominent and re-X-ray showed that the bony destruction extended laterally to the floor of the middle and external ear, while the right labyrinth gave a normal response to caloric stimulation (nystagmus 1° to the left after 45 seconds).

Accordingly the right mastoid area was explored on *20.7.55*, but revealed normal cells and a healthy sigmoid sinus: bone biopsy proved negative. In view of this the tympanic membrane was elevated from below and a biopsy taken from the mass in the hypotympanum. This was reported as "fibrous tissue covered by columnar epithelium, a moderate degree of round and polymorphonuclear cell infiltration is present. Some ganglion cells are incorporated in one area of the section. The appearances are those of a chronic inflammatory lesion and there are no signs of tumour".

Following this apparently negative result, examination of blood and urine



(a)

(b)

FIGS. 1a and 1b

was normal, as were X-rays of chest, pelvis and long bones. Observation was continued.

On *22.11.55* pale ballooning of soft tissues on the inner part of the floor of the right external meatus occluded the lower part of the drum, and by *10.4.56* this ballooning was more prominent and rather pink in colour.

On *21.4.56* she was admitted as an emergency, when examination revealed paralysis of right

VII. Facial, lower neurone type (whole face).

XI. Cranial: complete paresis right cord
palate
Spinal: sterno-mastoid,
with

VIII. Nystagmus
Past-pointing
Staggering.

as evidence that vestibular as well as auditory fibres of VIII were now involved.

On *23.4.56* a further biopsy from the right middle-ear was reported as "A mass of chronic inflammatory granulation tissue heavily infiltrated with plasma cells, hæmorrhagic in some places, and partially covered by stratified squamous epithelium, the features of a granulomatous polyp"; lumbar puncture and sternal puncture proved normal.

On *8.5.56* the right petrous bone was explored by the Lempert approach (Lempert 1937) but revealed no mass there, rather a defect inferior to the petrous, containing soft avascular fatty looking tissue.

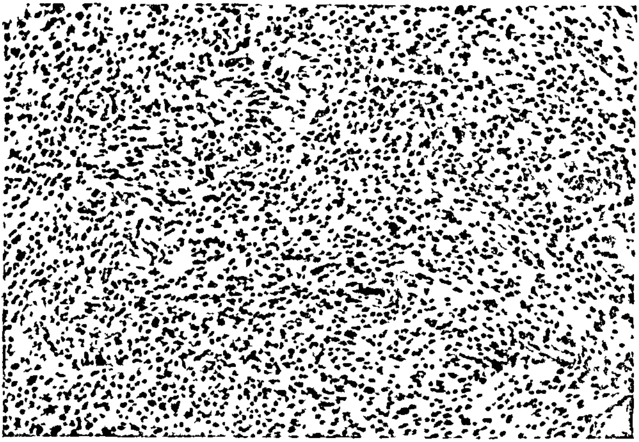
Biopsy from this material was reported as "much more highly cellular than in previous biopsies, and now raises the question of neoplasm—neurilemmoma is probably the most likely, but until tissue with a more clearly-defined and diagnostically distinctive pattern becomes available, this diagnosis must be regarded as *sub judice*. There is no evidence of lymphosarcoma or glomus jugulare tumour". Representative fields of this tissue are shown in Figs. *2a* and *2b*.

Following operation, fibrillation of the right margin of the tongue implicated the XIIth nerve, and contraction of the right pupil the sympathetic; in the absence of sensory loss and of muscle-wasting elsewhere, neurological opinion suggested a medullary lesion, the onset favouring a vascular cause.

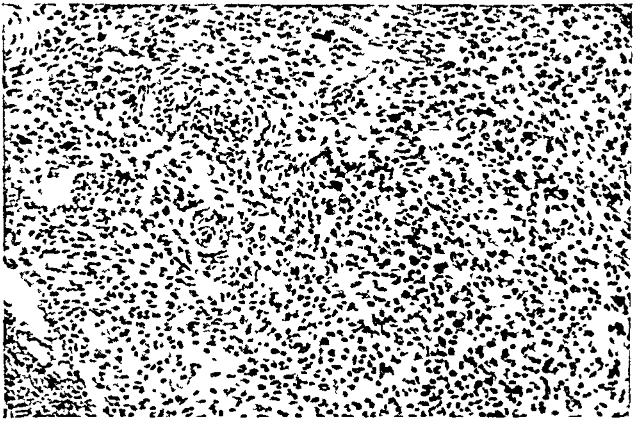
All these findings seemed to exclude glomus jugulare tumour and plasmacytoma, and to suggest neurilemmoma involving the petrous, and accordingly she was referred to Professor Dott for intracranial exploration.

On *5.7.56* extradural exploration of the right jugular foramen and petrous pyramid was performed through the post-auricular scar plus a transverse extension posteriorly. Exposure of the mastoid cavity, lateral sinus down to the jugular foramen, petrous pyramid and dura of middle and posterior fossae showed no tumour and only normal bony structure, and the operation was abandoned.

It was felt that there must be a neurilemmoma of the vagus nerve, arising on its extra-cranial portion just distal to its exit from the jugular foramen, with erosion upwards into the base of the medial petrous pyramid, and that nothing more could be done for this lesion at the present time, without undue risk which



(*a*)



(*b*)

Figs 2*a* and 2*b*

There is in some places a whorling of cells and a palisading of nuclei that suggests a neurilemmal neoplasm The tissue pattern is rather obscured by infiltrating lymphocytes, especially in Fig 1*a* H & E × 240 was unwarranted for a benign neoplasm which did not at present threaten her life.

Today (June, 1957) only slight weakness of right facial, palatal and lingual movement, but complete paresis of the right vocal cord remains, and she is well and active with no evidence of further extension.

Comment

Presenting as it did with vocal cord paresis, this case differs slightly from that of Love (1950), but the progressive deafness and facial palsy were also present. This presentation is reasonable evidence that the neurilemmoma originated in the vagus nerve, but it is difficult to exclude absolutely that it originated for instance on the glossopharyngeal, and involved the vagus secondarily. Penido *et al.* review 10 cases of tumour of the vagus nerve encountered in 20 years, of which 7 were neurilemmoma (1 malignant), 1 neurofibromatosis, and 2 chemodectoma, and in accessible situations such as the neck or mediastinum they advocate total excision and end to end repair rather than radiotherapy.

The differential diagnosis of such a bony defect of the petrous apex includes:

Glomus jugulare tumour as recorded by Heirons (1954) and Holesh (1955).

Plasmacytoma as recorded by Clarke (1954).

Cholesteatoma either aural or epidermoid by Pennybacker (1944), Jefferson and Smalley (1938) and Simpson (1957).

Xanthoma as recorded by Dingley (1952) and Dawes (1954).

Acoustic neurinoma seldom produces so much bony destruction before being very evident clinically, although Imrie (1930) does record a case with extensive destruction.

Other less common tumours such as meningioma, sarcoma, neuroepithelioma and carcinoma of nasopharynx or by metastasis from breast, bronchi, kidneys, prostate and thyroid are discussed by Lodge, Jones and Smith (1955).

Summary

A case of neurilemmoma of the Vagus presented as a Right Vocal Cord Paresis with X-ray evidence of destruction of the Right Petrous. The diagnosis, which required investigation of blood, urine, C.S.F. and bone-marrow, and repeated biopsy, is deduced and discussed.

Acknowledgments

I wish to thank Professor Norman M. Dott for his help in the intracranial investigation, and Dr. W. W. Park for the pathological detail and photomicrographs of this case.

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GENERAL NOTES

THE Editors are anxious to improve the clinical aspect of the Journal and would welcome more reports of interesting cases. These clinical records need not be at all elaborate and it would be unnecessary to go minutely into the ætiology or the literature of the subject. Senior Registrars have unique opportunities for seeing a large variety of cases, many with unusual features. It is in many ways regrettable that these should go unrecorded. It is hoped that all those in charge of clinics will help us in this matter and see that such cases are not overlooked.

OTOLARYNGOLOGY LECTURES 1957-58

THE following lectures, arranged jointly by the Royal College of Surgeons of England and the Institute of Laryngology and Otology, will be delivered in the Lecture Hall of the College, at 5.30 p.m.

1958
March 6th. Mr. H. Jackson Burrows. Disorders of the cervical spine.
May 1st. Professor W. D. M. Paton. The pharmacology of mucous membrane.
July 3rd. Mr. Norman C. Tanner. Haemorrhage from the Œsophagus.

INTERNATIONAL CONGRESS ON THE MODERN EDUCATIONAL TREATMENT OF DEAFNESS

AN International Congress on the Modern Educational Treatment of Deafness will be held at the University of Manchester from July 15th-23rd, 1958.

The Congress is being organized with the approval and support of the Council of the University, with financial help from the Ministry of Education, the National College of Teachers of the Deaf, and the Rathbone Trustees, and with the co-operation of the British Council and the Alexander Graham Bell Association for the Deaf, Inc., Washington, D.C., U.S.A.

The Congress is being planned to bring together educators, otolaryngologists, pædiatricians, medical officers, audiologists and educational psychologists, to consider and discuss the significance, to the future development of their work, of the results of modern research on problems of deafness in childhood.

Arrangements are being made to offer accommodation to members of the Congress in University Halls of Residence The cost of this type of accommodation is expected to be about £1 2s per day, including bed, breakfast and dinner. The University Refectory will be available for meals during each day

Forms of application for membership of the Congress will be supplied upon request, to Professor A W G Ewing, Department of Education of the Deaf, the University, Manchester 13, England

BRITISH ASSOCIATION OF OTOLARYNGOLOGISTS

Memorandum on its Formation, Activities and Objectives

In the early part of the war commencing in 1939 there was considerable difficulty in obtaining equipment for Ear Nose and Throat work in improvised hospitals Representations were made to the adviser to the Ministry of Health, but it was felt that individual requests were not likely to be so effective as concerted action Discussions took place as to the formation of some organization to give more effect to the requests of Otolaryngologists

In 1942 efforts were made by the President of the Sections of Otology and Laryngology, at that time Mr Ormerod and Mr Negus, to form a joint committee within the framework of the Royal Society of Medicine Discussions were held on this matter, and the President and Council had the views of representatives expressed to them It was felt, however, that the Society of Medicine could not take part in matters which were not of an academic nature

At the same time requests were made for proposals regarding education in Otolaryngology and these were formulated by a joint committee of the two Sections of Otology and Laryngology for the benefit of the Goodenough Committee This was an *ad hoc* Committee and it was still felt by many that a permanent organization was required A meeting was held, under the Chairmanship of Mr Colledge, and it was decided that a Society of Otolaryngology should be formed, with no connection with outside bodies

In 1942 the President and Council of the Royal College of Surgeons had issued an invitation for a representative to attend council meetings, and Mr Negus was deputed to accept this offer During his time of office the Council of the College of Surgeons put forward a suggestion that faculties should be formed within the College and that Otolaryngology should be one of them This however was not thought to be a good plan as freedom of action was preferred, and in consequence the British Association of Otolaryngologists has been in action since 1943

Amongst other activities, it has carried out a review of the Otolaryngological services of the Country and has put forward recommendations to the Ministry of Health It has also given information about the requirements of education in the specialty

Proposals were made by the Association to the Conjoint Examining Board and to the University of London, that there should be some means of testing the knowledge of candidates up for their Final Examination in Otolaryngology as is done at some other Universities, for instance Oxford and Sheffield. The extensive syllabus already existing was, however, thought by the two Boards to be a reason for not adding to the subjects of examination, and no steps were taken to implement the recommendations of the Association.

Many other matters have been considered by the Association, and their details have been presented in the regular Annual Reports.

The main objectives of the Association are, firstly to maintain a high standard of efficiency in Otolaryngology throughout the British Isles and secondly to safeguard, in every possible way, the interests of its Members.

BRITISH ASSOCIATION OF OTOLARYNGOLOGISTS

International Congress on the Modern Educational Treatment of Deafness

An International Congress on the Modern Educational Treatment of Deafness will be held at the University of Manchester from July 15th to 23rd, 1958.

The Congress is being organized with the approval and support of the Council of the University, with financial help from the Ministry of Education, the National College of Teachers of the Deaf, and the Rathbone Trustees, and with co-operation of the British Council and the Alexander Graham Bell Association for the Deaf, Inc., Washington, D.C., U.S.A.

The Congress is being planned to bring together educators, otolaryngologists, pædiatricians, medical officers, audiologists and educational psychologists, to consider and discuss the significance, to the future development of their work, of the results of modern research on problems of deafness in childhood.

Arrangements are being made to offer accommodation to members of the Congress in University Halls of Residence. The cost of this type of accommodation is expected to be about £1 2s. 0d. per day, including bed, breakfast and dinner. The University Refectory will be available for meals during each day.

Forms of application for membership of the Congress will be supplied upon request, to Professor A. W. G. Ewing, Department of Education of the Deaf, the University, Manchester 13, England.

The Fourth James Yearsley Lecture will be delivered by Mr. Francis McGuckin, F.R.C.S., at the Royal Society of Medicine, West Hall, on Friday, 7th March, 1958, at 5.15 p.m. The title of his lecture is "Recent Developments in Tympanoplastic Surgery".

General Notes

EXAMINING BOARD IN ENGLAND

BY THE

ROYAL COLLEGE OF PHYSICIANS OF LONDON

AND THE

ROYAL COLLEGE OF SURGEONS OF ENGLAND

DIPLOMA IN LARYNGOLOGY AND OTOLOGY

THE Secretary reports to the two Royal Colleges that at the First Part of the Examination held in December, 1957, 49 Candidates presented themselves, of whom 26 passed; and that at the Second Part of the Examination held in December, 1957, 33 Candidates presented themselves, of whom 21 passed.

The following 21 Candidates, having complied with the regulations of the Board, are eligible to receive the Diploma, viz:-

Name	Qualification	Address
*Abbey, Paul	M.B., S B. London (London)	40, Ashburn Place, S W 7
Ahmad, Rasheed	M.B., B.S Karachi	201, Bhurgri Road, Hyderabad, Sind
Alvi, Zaheerul Haque	M B , B S. Karachi	Sussex Throat and Ear Hospital, Brighton 1
Bihari, Julian	M B., B S London L.R.C P., F R C.S (U C.H.)	19, Fieldend Road, Streatham, S W 16.
Cartmill, Thomas	M.B , Ch B Glasgow	456, Victoria Road, Glasgow, S 2.
Coles, Robert Ross Adlard	M.B , B Ch Cambridge (St. Mary's)	"Cinderford", 39, St Mary's Avenue, Alverstoke, Gosport, Hants
Das, Bishnu Ram	M.B., B S Gauhati	Capra Guest House, 7-8, Brunswick Square, W C.1
D'Cruz, Manuel Joseph	M B., B.S. Gujarat	Whiston Hospital, Prescot, Lancs
Godden, John Lidington	B.M , B.Ch. Oxford (St Bart's.)	Flat 98, 6, Charterhouse Square, E C 1
Hall-Jones, John	M B , Ch.B New Zealand	E.N T Registrar, Royal Infirmary, Stoke-on-Trent
Horowitz, Martin	M B , B Ch. Cambridge (Manchester)	11, O'Leary Street, Orford, Warrington, Lancs.
Kapur, Yash Pal	M.B , B S. Punjab	136, Hampstead Road, Bristol 4
Khan, Ghulam Mustafa	M B., B.S Karachi	Barnet General Hospital, Wellhouse Lane, Barnet, Herts.
Kwok, Frank William	M.B , Ch B. New Zealand	109, St Mary's Mansions, St. Mary's Square, W 2.
Narain, Ishwar	M.B., B.S. Calcutta	c/o Royal National Ear, Nose and Throat Hospital, Grays Inn Road, W C.1.
Parvathiah, Nishanimath Basavalingaiah	M B., B S. Mysore	c/o Sri K C. Patre, Coffee Planter, Birur
Potter, Jeffery	M.B., Ch B. Manchester	17, Trent Boulevard, W. Bridgford, Notts

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* Examination passed previously.

Name	Qualification	Address
Prasad, Lakshmi Narayan ..	M.B., B.S. Patna ..	c/o Miss B. Bates, Colombo Plan Section, British Council, 65, Davies Street, W.1.
Prasad, Rajendra	M.B., B.S. Patna ..	Tufanganj. P.O. Soh Sarai, Dist. Patna, Bihar, India.
Robertson, Ian Peter Cameron	M.B., Ch.B. Glasgow	21, Maxwell Avenue, Westerton, Bearsden, Dunbartonshire.
Sardana, Dharmendra Singha	M.B., B.S. Lucknow ..	c/o Lloyds Bank, 67, Kingsway, W.C.2.

The Journal of Laryngology and Otology

(Founded in 1887 by MORRELL MACKENZIE *and* NORRIS WOLFENDEN*)*

March 1958

HISTOPATHOLOGICAL STUDIES OF THE OSSICLES IN CHRONIC OTITIS MEDIA

By M GRIPPAUDO (Rome)*†

IT has been recognized for many years that the ossicles may be diseased in chronic middle-ear suppuration Schwartze (1873) and Politzer (1884), in their classical writing on the pathology of chronic suppurative otitis media, emphasized the frequency of ossicular involvement Schulze (1904) and later Ferreri (1922) amplified these studies by histological investigations Since the advent of the modern era of antibiotic therapy and reconstructive surgery, this subject has, however, attracted but scant attention

Material and Methods

This paper is based on a study of 55 ossicles removed at the Royal National Throat, Nose and Ear Hospital, London, from cases of chronic suppurative otitis media over a period of eight years (1949-1957) It is the object of this study to consider in some detail the histological changes found in these ossicles and further, to attempt an assessment of their significance

The surgical source of the 55 ossicles studied is listed in Table I These ossicles were removed from 41 cases of chronic suppurative otitis

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* From the Department of Pathology, The Institute of Laryngology and Otology London

† Investigations carried out during the tenure of a research assistantship

TABLE I

	Radical Mastoidectomy	Epitympano-Mastoidectomy	Tympanoplasty	Total
Malleus	16	5	2	23
Incus	20	8	2	30
Stapes	1	0	1	2

OPERATIVE SOURCE OF THE 55 OSSICLES STUDIED

media, the relevant clinical data of which is presented in Table II. Most cases were characterized by persistent discharge and chronic attic disease. Case No. 10 was, however, defined as an "inactive otitis". As can be seen, Nos. 4, 9 and 40 were "reoperated cases".

The ossicles were fixed in 10 per cent. formol saline and sectioned according to the technique described by Friedmann (1955a). Thirty-one ossicles were serially sectioned.

Results

The histopathology of bone and mucosal changes will be considered under separate headings. For the sake of convenience these changes will be described according to the schema of Tables III and IV. It must, however, be appreciated that these classifications are somewhat arbitrary as all combinations and associations of lesions may be encountered. Unfortunately, no clinical observations are available, as to the state of disease or otherwise of these ossicles removed at surgery.

An understanding of the pathogenesis of ossicular changes in suppurative disease of the middle-ear cleft, demands familiarity with certain of their structural aspects. Gruber (1893), has emphasized the structural identity of ossicles and long bones. In fact one may regard ossicles as long bones in miniature.

The marrow cavities of the incus are more numerous than the malleus and in both are centrally located. The processes on the other hand, consist entirely of compact bone. The epithelium covering the ossicles is flattened, endothelial-like and firmly adherent to the underlying fibrous layer of the periosteum, thus constituting a fibro-mucosa. The blood supply of the ossicles has not yet been clearly defined. A branch of the stylomastoid or, according to Nager (1953), the anterior tympanic artery, passes to the incus through an attic mucosal fold, posterior to the malleus. It is thus particularly exposed to inflammatory processes. The vascularization of the malleus is more generous. In addition to receiving a twig from the anterior tympanic artery, it has also numerous vascular connections with the tympanic membrane. The stapes derives its blood supply from three

TABLE II

CLINICAL DATA AS TO SOURCE OF MATERIAL

No	Age	Sex	Clinical Diagnosis	Duration Disease in Years	Signs	Operation		Ossicles	Histopathology		
						Type	Findings		Bone Changes	Mucosal Changes	Epidermoid Cholesteatoma
1	20	F	R C O M	2	Cholesteatoma middle ear	Rd M	atoma	Malleus and incus	+	+	+ (attic)
2	37	M	R C O M	>10	Attic cholesteatoma	Rd M	ia	Incus	+	+	+ (attic)
3	49	M	L C O M	12	Meatal polyp	Rd M		Incus	—	+	—
4	9	M	L C O M	Not known	Eptm cavity discharge	Rd M	[illegible]	Incus	+	+	+ (attic)
5	44	M	L C O M	35	Attic disease	Rd M	ia	Stapes	+	+	+ (middle ear)
6	47	F	R C O M	40	Attic disease	Eptm	[illegible]	Malleus and incus	+	+	+ (attic)
7	32	F	R C O M	1	Attic disease	Eptm	[illegible]	Malleus	+	+	+ (middle ear)
8	39	F	L C O M	Not known	Attic disease	Eptm		Incus	—	—	—
9	11	M	R C O M	5	Eptm cavity discharge	Rd M	granu	Malleus and incus	+	+	+ (attic)
10	16	F	R and L C O M	>10	Inactive otitis	L Ty	[illegible]	Malleus and incus	+	+	+ (attic)
11	50	F	L C O M	45	Attic cholesteatoma	Rd M	[illegible]	Malleus and incus	+	+	+ (attic)
12	14	M	R and L C O M	7	Attic disease	L Rd M	[illegible]	Malleus	+	+	+ (attic)
13	17	F	R C O M	6	*Attic granulations*	Rd M	ia	*Incus*	+	+	+ (attic)
14	69	F	L C O M	>10	Attic cholesteatoma	Rd M	[illegible]	Malleus	+	+	+ (middle ear)
15	24	F	R C O M	20	Attic granulations	Rd M	tions	Malleus and incus	+	+	—
16	22	M	R and L C O M	17	[illegible]	L Ty	tions	Malleus and incus	+	+	+ (attic)
17	28	F	L C O M	10	ration	Rd M		Incus	+	+	—
18	22	M	R C O M	19	eatoma	Rd M	[illegible]	Incus	+	+	+ (attic)
19	24	F	R C O M	9		Rd M		Incus	+	+	—
20	53	M	R C O M	>10		Rd M		Incus	+	+	—
21	19	M	L C O M	9		Rd M	ear	Malleus	+	+	—
22	23	F	L C O M	>10	middle ear	Rd M	ear	Incus	—	+	—
23	30	M	R and L C O M	5	oration	L Rd M		Malleus and incus	+	+	—
24	41	M	R C O M	3	itions	Rd M	lle ear	Malleus and incus	+	+	—
25	43	F	L C O M	4		Eptm		Incus	+	+	—
26	57	M	R and L C O M	1	tion	R Eptm	eatoma	Malleus and incus	+	+	+ (attic)
27	51	M	R C O M	>10	a granula	Eptm		Incus	+	+	+ (attic)
28	54	M	L C O M	1	atoma	Rd M	Attic antral cholesteatoma	Malleus	+	+	+ (attic)
29	40	M	R and L C O M	20		Rd M	Attic cholesteatoma	Malleus and incus	+	+	—
30	46	F	R C O M	4		Eptm	Antrum cholesteatoma	Malleus	+	+	—
31	35	M	L C O M	>10		Eptm	Attic antral granulations	Malleus and incus	+	+	—
32	43	M	R C O M	15	foration	Rd M	Attic antral cholesteatoma	Malleus	+	+	— (attic)
33	12	M	L C O M	8	ration	Rd M	Granulations middle ear	Malleus and incus	+	+	—
34	20	F	L C O M	>10	ration	Eptm	No data	Incus	+	+	—
35	60	M	R C O M	Not known	foration	Rd M	Attic cholesteatoma	Malleus and incus	+	+	—
36	31	M	R C O M	8		Rd M	Granulations middle ear	Incus	+	+	—
37	46	F	L C O M	2		Rd M	Attic antral cholesteatoma	Malleus	+	+	+ (attic)
38	12	M	R C O M	7		Eptm	Attic antral cholesteatoma	Incus	+	+	+ (attic)
39	56	F	R and L C O M	Not known	ration	L Ty	Attic cholesteatoma	Stapes	+	+	—
40	19	M	L C O M	15	discharge	Rd M	Granulations middle ear	Malleus	—		—
41	48	F	R C O M	4		Rd M	Attic cholesteatoma	Incus	+	+	—

R — Right L — Left C O M — Chronic Otitis Media
Rd M = Radical Mastoidectomy Eptm — Epitympanomastoidectomy
Ty = Tympanoplasty

TABLE III

INCIDENCE AND NATURE OF BONE CHANGES.

	Changes		No. of Cases
Changes in bone spaces	1	Congestion and infiltration	33
	2	Granulation tissue	28
	3	Necrosis	1
	4	Vascular thrombosis	1
Bone formation		Lamellar and intramembraneous	30
Bone destruction	1	Absorption	28
	2	Necrosis	6
Sclerotic bone features			7

TABLE IV

INCIDENCE AND NATURE OF MUCOSAL CHANGES.

	Changes			No. of Cases
Epithelial layer	1	Keratinizing squamous		17
	2	Columnar secretory		14
	3	Cubical		5
Sub-Epithelial layer	1	Congestion and infiltration		6
	2	Granulation tissue	Simple	31
			Gland-like structures	5
			Cholesterol granulomata	3

sources, the superior and inferior tympanic arteries and a branch from the petrous artery. The crura and the thin part of the footplate, do not appear to contain blood vessels and according to Nager (1953) are nourished by diffusion from their mucosal network.

Bone Changes

As indicated in Table V, bone disease was encountered in 92 per cent. of the ossicles examined. Whilst the malleus and incus were involved with similar frequency, the extent and distribution of disease appeared greater in the latter. Furthermore, as indicated by Tumarkin (1950), Dingley and

TABLE V

INCIDENCE OF BONE CHANGES IN THE OSSICLES.

	Normal	Diseased	Total
Malleus ..	1	22	23
Incus .. .	3	27	30
Stapes . ..	0	2	2

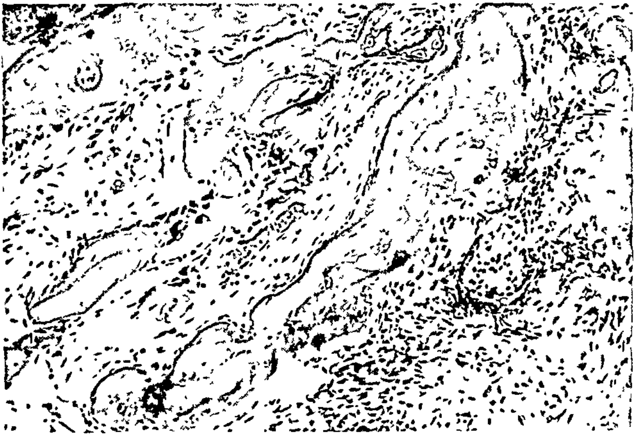

FIG 1

Section of incus showing distended bone spaces obliterated by granulation tissue × 330 Haematoxylin and eosin

Zwiefach (1954), the presence of attic cholesteatoma seemed to be associated with more constant and severe bone changes in the ossicles.

The incidence and nature of these bone changes is listed in Table III. Changes in bone spaces were very common. The most frequent were congestion and infiltration by lymphocytes and plasmacytes, which were sometimes disposed as perivascular cuffs. In some cases these infiltrations constituted the sole abnormality. Bone spaces were sometimes distorted and in 28 cases obliterated by granulation tissue (Fig. 1). Marrow necrosis was found in one case, presumably due to vascular thrombosis (Fig. 2). Vascular thrombosis was, however, seen in one case only (Fig. 3).

Two types of bone formation were observed—intramembraneous and

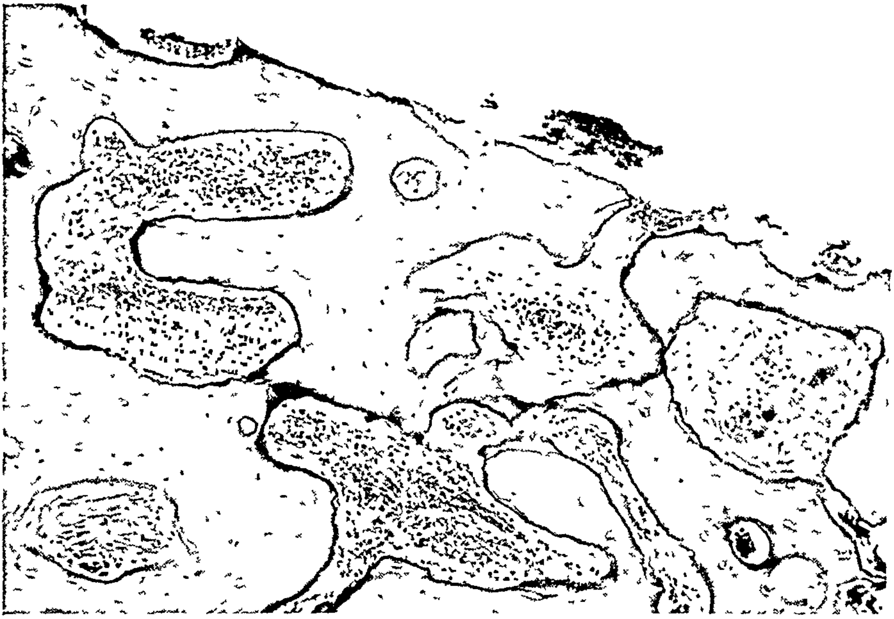

FIG 2

Section of incus showing necrosis of marrow in the distended bone spaces × 220 Haematoxylin and eosin

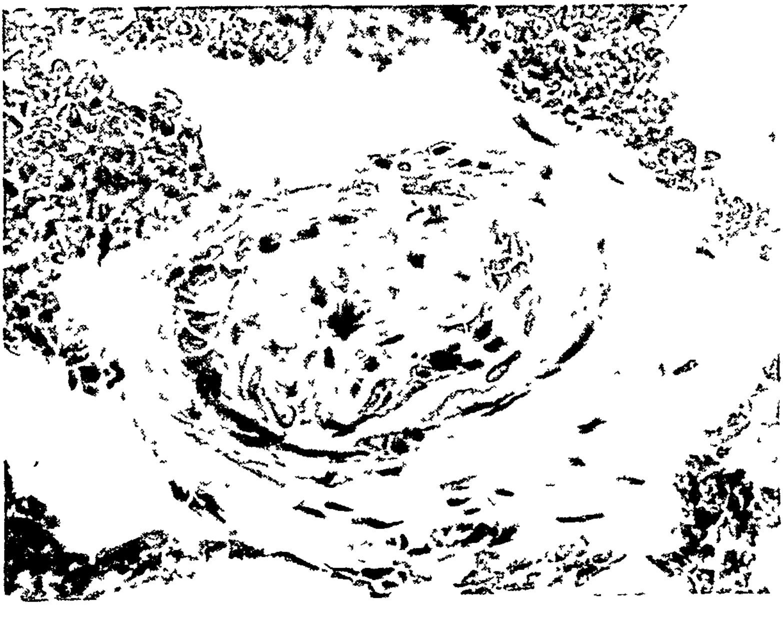

FIG 3.

Section of incus showing vascular thrombosis × 1150 Haematoxylin and eosin lamellar Intramembraneous osteogenesis (Fig 4), is less common and arises from the connective tissue replacing destroyed bone Lamellar osteogenesis, however, originates from periosteal osteoblasts or those lining bone spaces The former may result in the formation of osteophytic projections and an overall increase in the size of the ossicles The latter can produce obliteration of bone spaces and osteosclerosis Newly formed bone presents a distinctive appearance, being eosinophilic and in the case of lamellar bone, subsequently exhibiting irregular dark blue lines of apposition, indicating previous bone absorption and bone reconstruction

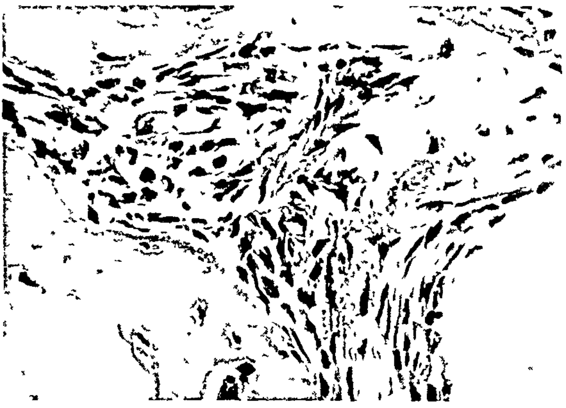

FIG 4

Section of incus showing new bone formation × 1100 Haematoxylin and eosin

Bone absorption was frequently found and was due to osteoclasia, vascular erosion, halisteresis, or any combination of these three Osteoclasia (Fig 5) was particularly observed in cases of recurrent acute exacerbations Extensive bone necrosis was not found in our material Patchy necrotic areas and small sequestra, occasionally the site of intensive calcium deposition, were sometimes seen It must, however, be emphasized that in 21 cases, the long process of incus could not be found and it was difficult to decide whether it had been destroyed by infection, or necrosis due to vascular thrombosis In most cases the articular surfaces of the ossicles exhibited no abnormality In five cases, however, erosion of variable extent was found

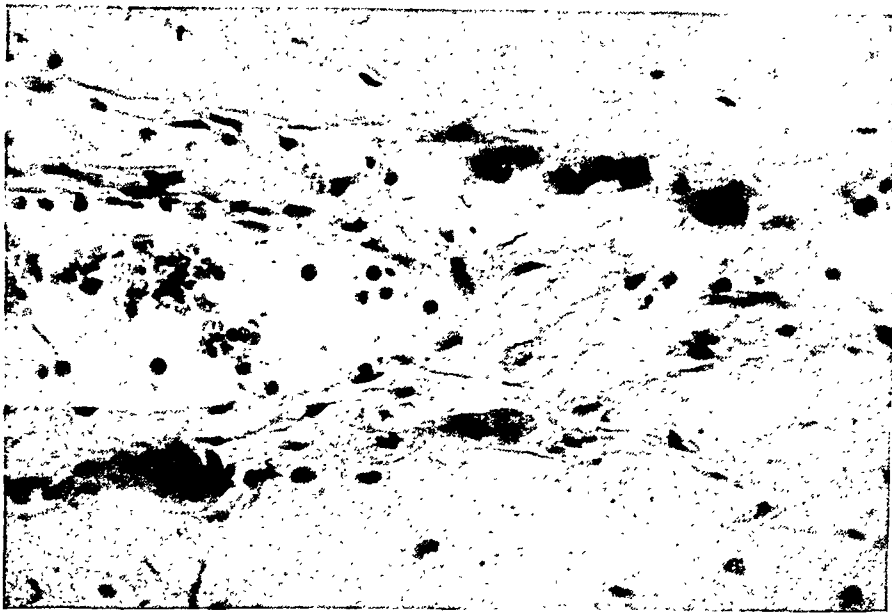

FIG. 5.

Section of malleus showing osteoclastic activity. × 750. Haematoxylin and eosin.

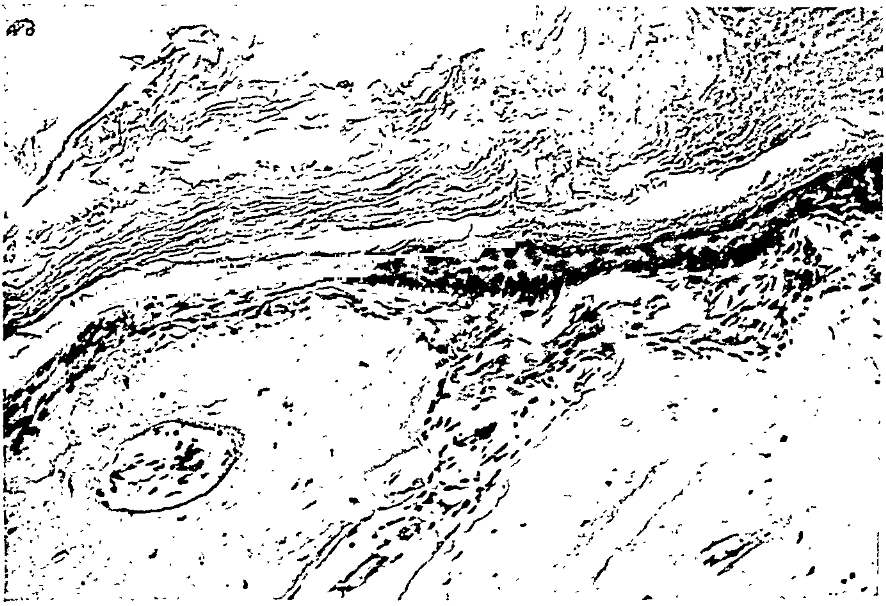

FIG. 6.

Section of malleus showing lining of epidermoid cholesteatoma covering the surface of the bone. × 340. Haematoxylin and eosin.

granulation tissue. Cholesterol granulomata were found in the subepithelial tissues in three cases. As explained by Friedmann (1956) these consist of cholesterol clefts embedded in fibrous granulation tissue surrounded by foreign-body giant cells.

Discussion

The bone changes reviewed above are more or less manifestations of chronic osteomyelitis. Schulze (1904) and Ferreri (1922), among other writers in the pre-antibiotic era, arrived at similar conclusions. This was confirmed by Friedmann (1956) who, on the basis of human and experimental findings, has emphasized that the fundamental pathology of otitis media has not changed in the antibiotic era.

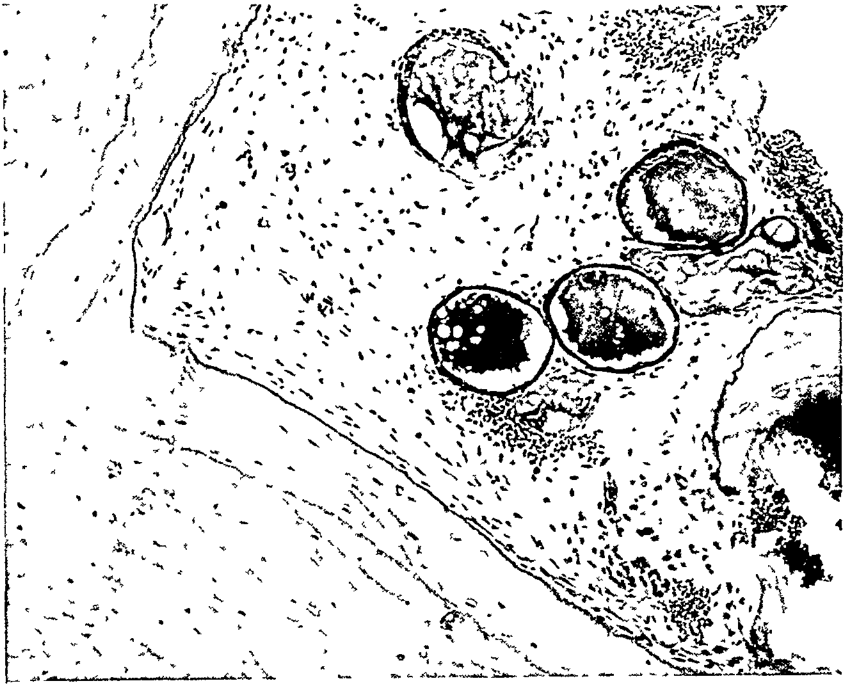

FIG 8

Section of incus showing sclerotic bone and gland-like structures in some granulation tissue × 270 Haematoxylin and eosin

The almost invariable involvement, by histological criteria, of ossicles in chronic suppurative otitis media, has already been emphasized. This contrasts with the views expressed by authors such as Ramadier and Pialoux (1951) who, on the basis of clinical observations, claim that normal bone is found in 20 per cent. of cases.

In accordance with the findings of Gruber (1893), Ferreri (1922) and

Jackson (1945), the malleus and incus were involved in the material studied in equal proportion. Schwartze (1873) maintained that the malleus is more often affected than the incus, whilst Grunert (1892) among others, held the opposite views. The greater extent of disease usually found in the incus, is probably merely a reflection of its poorer vascularity and cancellous nature. The rarity of removal of the stapes, makes it difficult to assess the frequency of its involvement. Most authors, as Goto (1956) agree that the stapes is not commonly affected, possibly due to its structure and well-developed blood supply. In the two specimens studied in this survey, true osteomyelitic changes [illegible] logically, but the footplates were normal (Fig. 9).

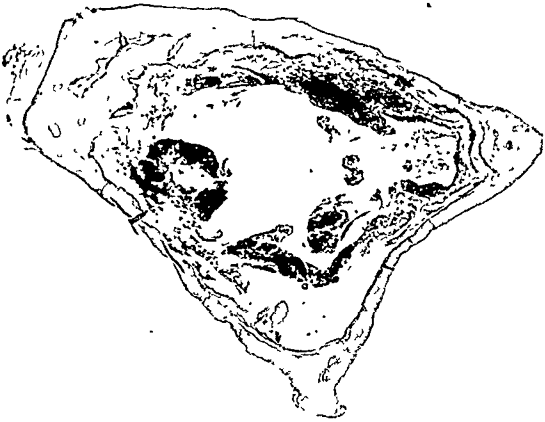

FIG 9

Section of stapes containing purulent granulation tissue × 30 [illegible] and eosin

The mucosal changes above described, likewise are [illegible] chronic inflammation. Reversal of the flattened [illegible] epithelial lining, to the embryonic respiratory type in [illegible] been described by Friedmann (1956) in mastoid cells.

Equally squamous epithelium was a common fi[illegible] ossicles in chronic suppurative disease and formed the [illegible] moid cholesteatomata. The replacement of granulation [illegible] can result in an adhesive otitis with the subsequent imp[illegible]

Carnevale-Ricci (1955), Del Bo (1956), Ormerod and McLay (1957) have emphasized that the first essential of plastic treatment of chronic suppurative otitis media is the cure of underlying disease. The findings described above suggest the possibility that the retention of diseased ossicles may well prejudice the success of reconstructive surgery.

Summary

The paper presents a histological analysis of 55 ossicles surgically removed from 41 patients with chronic suppurative otitis media, at the Royal National Throat, Nose and Ear Hospital, London, over a period of eight years (1949-57). Histologically normal ossicles were only seen in four cases.

The malleus and incus were affected in the same number of instances, the incus more obviously, by various bone and mucosal changes. The latter consisted essentially of epithelial changes and granulation tissue formation, the former of new-bone formation and inflammatory processes in bone spaces. The intensity of these changes was more marked in the presence of attic epidermoid cholesteatoma.

The present studies confirm the findings of earlier authors and indicate that the advent of the antibiotic era has not been associated with an alteration of the fundamental pathology of chronic suppurative otitis media.

Attention is drawn to the possibility that diseased ossicles may prejudice the results of reconstructive surgery.

Acknowledgments

The author wishes to thank sincerely his chief, Professor G. Ferreri, for leave of absence and for every encouragement; Professor F. C. Ormerod for arranging this fruitful visit to the Institute of Laryngology and Otology in London; to Dr. I. Friedmann for suggesting the present investigation and his interest and help in the preparation of the paper; Dr. S. S. Epstein, for reading the manuscript and helpful advice; Miss V. Shepherd for the large number of sections; to Mr. D. Connolly for the excellent photographs and to Miss T. Hurlston and Miss S. Collins for typing and re-typing the manuscript. Finally he would like to express his grateful thanks to the surgical staff of The Royal National Throat, Nose and Ear Hospital, London, for permission to study the clinical notes of their patients; and Mr. J. H. Young, House Governor, for his kindness and help.

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THE TREATMENT OF MÉNIÈRE'S DISEASE WITH ACETAZOLAMIDE

By GY. VARGA (Budapest)*

(Preliminary publication)

THE immediate cause of Ménière's disease is undoubtedly the increase of the endolymph in the inner ear, the cause of which is again supposed to be vegetative vasoneurosis.

The principal symptoms of the disease are: attacks of vertigo, the diminution of the vestibular erethism, tinnitus and periodical hardness of hearing. The fixing of the correct diagnosis is essential. For that purpose the examination of the hardness of hearing with the audiometer gives the necessary directions. Often one must experiment with treatment, but already it is certain that a diet poor in salt and fluid and the use of diuretics have not proved successful. The result is the same when sodium-chloride is substituted for potassium chloride.

In consequence of the results obtained in cases of glaucoma, we have tried applying acetazolamide in case of properly diagnosed Ménière's disease. This drug diminishes the intra-ocular tension. It also has a specific effect. It exerts an influence on the nervous system, as is shown by the results gained in cases of epilepsy. Under its influence as a result of the paralysing of the carbonic anhydrase enzyme the excretion of sodium, potassium and bicarbonate increases. We have examined the increasing excretion of sodium and potassium with Zeiss's Flame Photometer. The balance of acid-base changes, slight acidosis and the resorption of the œdemas takes place. The drug was put on the market under the name Diamox.

Until now we have given Diamox to five patients afflicted with Ménière's disease.

1. R.M., 40 years old, ill for eight years. He has vertigo of vestibular origin on the left side. Nausea and vomiting is frequent. On the left side the caloric erethism ceased to exist. On this side the hearing diminished by degrees, the hearing of whispered voice was 10 cm. At the audiometrical examination the average air conduction was 80 db, the bone conduction was not measurable. Regression (Fowler) was traceable, which

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also shows the injury of the nerve ending mechanism After the examinations we prescribed a diet, which contained daily ½ g sodium chloride and 1 lit fluid After the diet, on the third day the patient received on an empty stomach 500 Diamox and later, on the same day further 3×250 mg. of it On the following days, for ten days the patient received 4×, later on 3× 250 mg of Diamox Unpleasant secondary effects were not once observed On the eighth day we increased the salt content of the diet, on the eleventh day we introduced the normal menu In the meantime the objective and subjective symptoms improved by degrees Vertigo and the other complaints of the vestibular apparatus ceased Hearing of whispered voice improved from 10 cm to 4 m The air conducting audiogram showed an increasing of 35 db The bone conducting audiogram also increased, but the regression didn't change We experienced the same in the case of other patients, too The excretion of sodium was before the treatment of 1,120 mg and that of potassium 1,860 mg As a result of the diuretic drug the maximum of sodium excretion was daily 1,470 mg and that of potassium 3,760 mg The amount of urine increased nearly threefold (Fig 1)

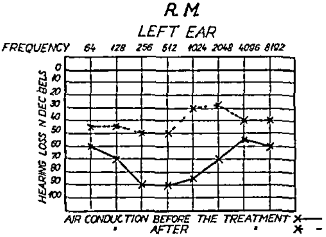

Fig 1

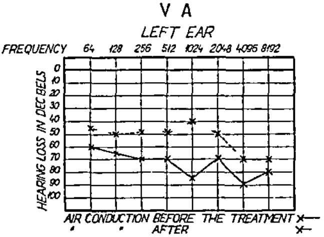

Fig 2

2 V A , 61 years old For one year he has complained of vertigo of rotatory character His left ear is buzzing, and on this side he does not hear well (ac whispered voice) After entering the hospital we observed acute attacks with nystagmus of secondary degree and with vomiting After the diet he was given 1,250 mg Diamox, in consequence of it the vertigo ceased and the buzzing of the ear diminished After having Diamox for four days (4×, later on 3× 250 mg) there was no vestibular symptom, hearing became 1, 5 m (ac whispered voice) The audiometrical examination showed an increase of the air conduction with 25 db Before the treatment the sodium-emptying was 1,720 mg and this of potassium 830 mg After the treatment the maximal content of sodium in the urine was daily 3 850 mg and that of potassium 4,000 mg The amount of urine increased in this case also threefold (Fig 2)

3. C.E., 52 years old. Three years ago he became aware of buzzing in the left ear and that he doesn't hear well. Soon after he felt himself uncertain and later he had attacks of vertigo. At present he hears in his left ear 40 cm. spoken voice. After the treatment the vertigo and buzzing in the ear ceased, but the hearing didn't change only when he called for hospitalization. In result of the treatment his hearing increased in case of whispered voice to 3, 5 m., the air conducting hearing with 25 db. Before the dietetic treatment the emptying of sodium was 1,720 mg., and that of potassium 1,321 mg. After the dosing of Diamox the maximal content of sodium in the urine daily was 3,000 mg. and that of potassium 3,900 mg. The amount of urine was more than twofold of the amount during the dietetic treatment (Fig. 3).

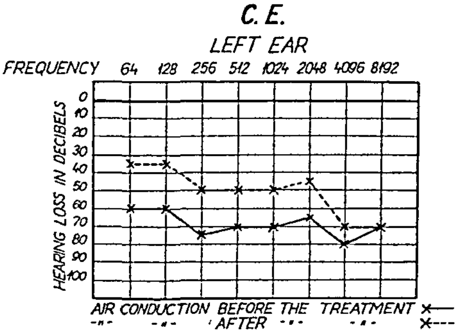

FIG. 3.

4. Cs. J., 33 years old. For half a year he had typical attacks of vertigo. For three days his left ear is strongly buzzing, the hardness of hearing increases by degrees on the left ear (ac. whispered voice). He received Diamox for seven days in the usual way. In result of the treatment the vertigo ceased, the buzzing in the ear diminished, but the hearing hardly changed (0, 1 m. whispered voice). The audiogram practically didn't alter. The maximum of sodium content in the urine was before the treatment 2,340 mg., that of potassium 1,640 mg., and after the treatment 3,900 mg., resp. 4,500 mg. The amount of urine increased twofold.

5. P.L., 37 years old. For 14 years he has typical attacks of vertigo of rotatory character. His right ear is buzzing, he hears very badly (ac. whispered voice). After the treatment for 10 days in the usual way with Diamox the vertigo and buzzing in the ear ceased, but the hearing didn't improve. The audiogram increased also only with 10 db. The maximum of sodium-emptying was 8,400 mg., that of potassium 4,200 mg. compared to the control of 3,000 mg., resp. 1,600 mg.

We have proved the effect of acetazolamide until now on only five patients. It is beyond doubt that the number of the patients and the *time of observation (after a control of two months the result didn't* change) is not enough for the exact recognition of the effect of the drug. We observed that in the case of three patients the symptoms greatly ceased after the treatment with Diamox compared to other drugs. The symptoms of two other patients also ceased partly, but we didn't observe appreciable increase of hearing. The results were not in relation to the duration of the disease. In connection with the dose of the drug it appeared that the lesion of the nerves is in some degree reversible. It is essential that the immediate cause, hydrops, shall be eased. This aim we gained with the emptying of sodium and potassium. The neural effect is also essential. The after-treatment consists of the dosing of various sedatives, of drugs against the vasomotor irregularities, of the restoring of the normal regimen, of the possible eliminating of all disturbing conditions. The correct diagnosis is very essential. We continue the experiments, because it seems that we may attain very good results with Diamox.

I am very grateful to Dr. Pál Kertai, demonstrator, and Dr. Otto Ribari, assistant, for their valuable collaboration.

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A REVIEW OF TUMOURS OF THE GLOMUS JUGULARE WITH REPORTS OF THREE FURTHER CASES

By I. C. SIMPSON and RUSSELL DALLACHY (Glasgow)*

DURING the years prior to 1945, there appeared in the literature a growing number of reports of unusual vascular tumours of the middle ear which were often associated with extensive destruction of the petrous temporal bone and with cranial nerve palsies. Biopsy specimens were usually described as of vascular origin. In 1945, Rosenwasser reported one of these tumours, described and illustrated its microscopic appearances which were similar to those of a carotid body tumour, and suggested that it might have its origin in the jugular glomera which had been described by Guild (1941). Since Rosenwasser's original article many examples of this tumour have been reported. A lesser number was recorded before 1945. In the present paper, probable or definite cases of glomus jugulare tumour of the middle ear since 1889 are reviewed; the pathology, clinical features and treatment are described; and three more cases are reported. The first of these patients has been under observation for 19 years and the second for 17 years. In the third patient, the tumour became apparent while the case was under investigation for deafness.

Origin and Site of Tumours of the Glomus Jugulare

The *glomus jugulare* is the name given to a small structure which is normally found in the human temporal bone; occasionally two or more glomera are present. Each glomus is flattened, ovoid and measures about 0·5×0·5×0·25 mm. (Guild, 1941). According to Lattes and Waltner (1949), the glomera in the temporal bone are found in the vicinity of the middle ear in the following sites: (i) in the adventitia of the jugular bulb just beneath the floor of the middle ear; (ii) in the bony walls of the tympanic canals which transmit the tympanic branches of the glossopharyngeal and vagus nerves; and (iii) in the bone of the promontorium close to the mucosal lining of the middle ear.

On microscopic examination, each glomus jugulare consists of solid masses of "epithelioid" glomus cells These masses show moderate variation in size They are situated between blood vessels which are usually capillaries, but which are occasionally larger and have smooth muscle fibres in their walls There are numerous nerve fibres in the normal glomera, these fibres are mainly sensory and come from the glossopharyngeal nerve The arterial blood supply is from the ascending pharyngeal artery through the inferior tympanic branch Thus the innervation and blood supply of the jugular glomera are derived from the same sources as those which supply the carotid body

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The histological appearance of the glomus jugulare is similar to that of the glomus caroticum (carotid body), situated at the bifurcation of the common carotid artery, and of the glomus aorticum (aortic body) which is a collection of tissue in the wall of the aorta in the ascending thoracic part and in the arch The carotid body and aortic body are chemoreceptors Their afferent nerve fibres are bound up with the vagus, in addition, the carotid body has an efferent twig which joins the glossopharyngeal nerve Nerve fibres are numerous The nerve endings in the "epithelioid" glomus cells are not sensitive to stretching stimuli but respond to chemical changes in the blood flowing through the sinusoids (Heymans and Bouckaert, 1939) It is believed that the glomus jugulare likewise functions as a chemoreceptor, although its influence is restricted to a smaller circulatory region (Guild, 1941)

Other structures which contain tissue which is histologically similar to that of the glomus jugulare, glomus caroticum and glomus aorticum are the ciliary ganglion, the ganglion nodosum of the vagus nerve and (probably) collections of cells in the walls of various arteries throughout the body but especially those in the lower limbs (Smetana and Scott, 1951)

Literature and Nomenclature

Cases of Tumour of the Glomus Jugulare reported prior to 1945 were frequently regarded as of vascular origin, often arising from the temporal bone The names given to the growth included hæmangioma, hæmangioendothelioma, fibrohæmangioma of petrous bone, hæmangiomatous polyp and angiosarcoma (see Table I) The fallacy of diagnosing a vascular tumour as a true tumour of blood vessels, especially if it shows evidence of associated proliferation of mesenchymal tissues, is now well known The fact that many of the glomus jugulare tumours recorded before 1945 were called angiomas of some kind was probably the result of failure to distinguish between the "epithelioid" chemoreceptor cells which are characteristic of these tumours and the vasoformative connective tissue cells ("pericytes") which accompany the vessels and sinusoids in angiomas and angiomatoid malformations

Another group of mistaken diagnoses included adenoma of a sebaceous

TABLE I

UNUSUAL VASCULAR TUMOURS OF MIDDLE EAR REPORTED PRIOR TO ROSENWASSER (1945)

Year	Authors	No of Cases	Original Diagnosis	Results of Review
1889	Wagenhauser	1	Angioma	Not accepted (Berg)
1896	Leutert	1	Endothelioma Lymphaticum	Probable case (Berg)
1897	Tervaert and de Jong	1	Angiosarcoma	Not accepted (Berg)
1897	Brieger	1	Angioma	Not accepted (Berg)
1897	Milligan	1	Angioendothelioma or angiosarcoma	Not accepted (Berg)
1899	Haug	1	Endothelial carcinoma	Probable case (Berg)
1904	Manasse	1	Endothelioma	Probable case (Winship *et al*)
1906	Beck	1	Hæmangioendothelioma	Probable case (Winship *et al*); doubtful case (Le Compte)
1907	Bryant	1	?	Probable case (Winship *et al*), doubtful (authors)
1908	Bentzen	1	Angioendothelioma or angiosarcoma	Not accepted by Berg
1909	Urbantschitsch, E	1	Angiosarcoma	Probable (Winship *et al*), not accepted by Gaffney and Le Compte
1909	Von Hammerschlag	1	Angioma	Not accepted (Berg)
1912	Stein	1	Lymphangiomatous tumour	Probable case (Gaffney)
1912	Jack	1	No path diagnosis made	Probable case (authors)
1912	Froning	1	Endothelioma hæmorrhagica	Not accepted (Berg)
1918	Ruttin	1	Angioma	Not accepted (Berg)
1919	Bilancioni	1	Sarcoma	Probable case (Winship *et al* and authors)
1919	Calabresi	1	Fibro myxo hæmangio endothelioma	Not accepted by Le Compte
1922	Mayer	1	Angioma	Not accepted (Berg)
1922	Gavello	1	Angioma	Not accepted (Berg)
1922	Layera	1	Angioendothelioma or angiosarcoma	Not accepted (Berg)
1923	Peters	1	Angioma	Not accepted (Berg)
1923	Piazza	1	Hæmangioendothelioma of external auditory meatus	Quoted by Berg—probable case (authors)
1925	Fischer	1	Angioma	Not accepted by Le Compte
1925	Cleminson	1	Endothelioma	Not accepted (authors).
1926	Marx	1	Hæmangioendothelioma	Not proven (Le Compte)
1926	Torrini	1	Angioendothelioma or angiosarcoma	Not accepted (Berg)
1927	Krepuska	1	Angiosarcoma	Accepted by Berg and Gaffney
1927	Weill and Bernheim	1	Angioma	Not accepted (Berg)
1928	Leidler and Sternberg	1	?	Accepted by Bartels and Gaffney Included in Bartels' series
1928	Richman	1	Angioma	Not accepted (Berg)
1929	Guisez and Richez	1		Probable case (Winship *et al* and authors)
1929	Specht and Völker	1	Hæmangioendothelioma	Not accepted by Le Compte
1930	Fraser	2	Case 10—sebaceous adenoma	Probable case (Winship *et al* and authors)
			Case 11—endothelioma	Probable case (Winship *et al* and authors)
1930	Dundas Grant	1	Endothelioma	Probable case (Berg)
1930	Jones	1	Hæmangioma	Not accepted by Le Compte
1930	Bronzini	1	Hæmangioendothelioma	Probable case (Winship *et al* not accepted by Le Compte)
1931	Fowler	1	Angiomatous polyp	Not accepted (Berg)
1931	Robinson	1	"Malignant tumour"	Probable case (Winship *et al* and authors)
1932	Zander	1	Angioma	Not accepted (Berg)
1932	Colledge	1		Probable case (Gaffney)
1932	Goekoop	3		Accepted by Gaffney, Bartels and Winship *et al* (Included in Bartels' series)
1933	Grossmann and Leidler	1	Angioendothelioma or angiosarcoma	Not accepted (Berg).
1933	Caussé	1	Angioma	Not accepted (authors)
1933	Bourgeois	2	Angioma	Not proven (Berg)
1933	Bergera and Bergera	1	Angioma	Not proven (Berg)
1933	Ormerod	1	Angiosarcoma	Accepted by Winship *et al* probable case (authors)
1934	Seiffert	1	Endothelioma	Accepted by Berg and Gaffney.
1934	Markow and Rosenhaus	1	Angioma	Not proven (Berg)
1934	Sullivan	1	Hæmangioendothelioma	Not accepted by Le Compte Probably a sarcoma (authors)
1935	Schall	2	Hæmangioendothelioma	Not accepted by either Le Compte or Berg
1935	Thorell	1		Probable case (Winship *et al* and authors)
1936	Jeschek	1	Hæmangioendothelioma	Accepted by Berg and Gaffney
1937	Altmann	1		Accepted by Berg
1937	Lubbers	1	Metastatic carotid body tumour	Accepted by Gaffney and Bartels (Included in Bartels' series)
1938	Urbantschitsch, E	1	Angiosarcoma	Probable case (Gaffney)
1938	Löwy	1	Epithelial tumour of alveolar structure	Probable case (Berg)
1939	Mackenzie	1	Hæmangioma of petrous bone	No biopsy—probable case (authors)

TABLE I—*cont*

Year	Authors	No of Cases	Original Diagnosis	Results of Review
1939	Hampton and Sampson	2	Angioma	Not proven (Berg)
1939	Falk	1		Accepted by Berg (his Case 1)
1939	Scott	1	Hæmangioendothelioma	Not proven (Le Compte) no photomicrograph
1939	Irgens	1	Hæmangioendothelioma	Not accepted (authors)
1943	Figi and Hempstead	7	Angioendothelioma	Not accepted on review by Figi
1943	Horn and Stout	2	Granular cell myoblastoma	Accepted by Berg and Gaffney
1943	Lorenz and Pogany	1	Angioendothelioma	Not proven (Berg)
1943	Carrea and Insausti	1	Angioma	Not proven (authors)
1943	Weaver	1	Angioma	Not proven (Berg)
1944	Capps	2	Hæmangioendothelioma	Accepted
1945	Dixon	1	Hæmangioma	Probable case (Gaffney)

gland (Fraser, 1930) and "epithelial tumour of alveolar structure" (Lowy, 1938). The occurrence of rounded groups, or trabeculae, or irregularly shaped masses of "epithelioid" glomus cells can give rise to appearances similar to those of abnormal or aberrant glandular structures.

Two cases of "granular cell myoblastoma" of intratympanic origin were reported by Horn and Stout (1943), but according to Capps (1952), this diagnosis was subsequently withdrawn by the original authors who agreed that the lesions which they had described were in fact tumours arising in the glomus jugulare. Khanolkar (1947) reported one example of "granular cell myoblastoma" of the middle ear in a paper in which he described patients who had tumours of this type in various parts of the body. He does not illustrate the tumour in the middle ear but it seems likely from the histological description that it was a tumour of the glomus jugulare. Willis (1953) considers that the term "granular cell myoblastoma" should be discarded, since tumours which have been described under that name may arise in tissues which do not contain embryonic muscle fibres.

Table I contains 14 cases which are acceptable as examples of tumours of the glomus jugulare and 19 which are regarded as probable cases, and 13 which are possible cases but are not proven. In addition, there are 50 cases of unusual vascular tumours which showed some clinical or pathological features of glomus jugulare tumours, but which are not classified as either "accepted" or "probable" because of incomplete descriptions. Many of the older cases have no photomicrographs of the tumour. A case has been regarded as acceptable if a satisfactory photomicrograph is available or if there is an adequate description of the characteristic structure of the tumour. It seems likely that many more tumours of the glomus jugulare must have occurred but these would not be reported because the significance of their structure was not understood. In the first of the three patients described in the present paper, two biopsies were taken from a tumour which presented in the external auditory canal and both specimens were reported in the first instance as fibrohæmangioma. The correct histological diagnosis was made only after the sections had been re-examined in the light of work published after the second biopsy was obtained. The second case was diagnosed on review of the sections when the patient was seen again while this paper was in preparation.

Cases of Tumour of the Glomus Jugulare reported during and after 1945. It has been pointed out at the beginning of this paper that in 1945 Rosenwasser described an intratympanic tumour which was similar in its histological structure to the "potato tumour" which occasionally arises in the carotid body. He also suggested that the tumour originated in the glomus jugulare. It should be noted that in 1937 (eight years before the publication of Rosenwasser's paper), Lubbers in Holland drew attention to the similarity in histological structure between a vascular tumour of the middle ear and the carotid body tumour. He was not aware of the presence and histological appearance of the normal glomus jugulare and consequently was not able to make any contribution to the theory of the pathogenesis of the growth which he described.

Following Rosenwasser's original paper in 1945 the number of recorded cases has increased steadily (Table II). Several authors re-examined old sections and reviewed cases of unusual vascular tumours of the middle ear which had occurred or had been recorded in the literature before 1945; a

TABLE II

GLOMUS JUGULARE TUMOURS OF MIDDLE EAR.
CASES REPORTED AS SUCH SINCE ROSENWASSER (1945).

Year	Authors	No. of Cases	Cases Accepted as Glomus Jugulare Tumours of Middle Ear	Comment
1945	Rosenwasser	1	Accepted	Reported as neoplasm of carotid body structure in middle ear.
1946	Matusevich	1		Probable case (Gaffney).
1947	Kipkie	1	Accepted	
1947	Le Compte, Sommers and Lathrop	1	Accepted	
1947	Proctor and Lindsay	1		Case 1, diagnosed as "meningothelial meningioma" — probable case (authors).
1947	Khanolkar	1		Case 6—probable case (authors).
1947	Eggston and Wolff	1	Accepted	
1948	Czurda and Köhlmeier	2	Accepted	
1948	Winship, Klopp and Jenkins	2	Accepted	
1948	Revilla (quoted by Bickerstaff and Howell)	1	Accepted (Bickerstaff and Howell)	
1949	Fuller	1	Accepted	
1949	Lattes and Waltner	8	Accepted	
1949	Lundgren	4	Accepted (Berg)	Included one case (Falk, 1939).
1949	Zettergren	1	Accepted (Berg)	
1949	Bartels	6	Accepted	Also reviewed 3 cases of Goekoop (1932), 1 case of Lubbers (1937), 1 case of Leidler and Sternberg (1928), 2 cases of Czurda and Kohlmeier (1948).
1950	De Lisa	1	Accepted	
1950	Graf	8	1 case (Case 4) accepted	Remaining cases probable.
1951	Lewis and Grant	2	Accepted (by Gaffney)	
1951	Poppen and Riemenschneider	2	1 case (Case 1) accepted by Gaffney	Case 2 not proven.
1951	Dockerty, Love and Patton	1	Accepted	
1951	Tamari, McMahon and Bergendahl	2	Accepted	
1951	Gibson and Proud	1	Accepted	
1951	Brown, K. B.	1	Accepted (Rosenwasser, 1952)	
1951	Weille and Lane	3	Accepted	
1951	Alexander, Beamer and Williams	1	Accepted	

TABLE II—*cont*

Year	Authors	No of Cases	Cases Accepted as Glomus Jugulare Tumours of Middle Ear	Comment
1951	Winship and Louzan	1	Accepted	
1951	Mattick and Mattick	2	Accepted	
1951	Hoople	3	Accepted (Rosenwasser 1952)	Unpublished Reported in discussion in Weille and Lattes paper
1951	Walsh	4	Accepted (Rosenwasser 1952)	
1951	Miller	1	Accepted (Rosenwasser 1952)	
1952	Cleary	1	Accepted	
195	Mattick and Burke	3	Accepted	Cases 1 and 2 included in Mattick and Mattick (1951)
1952	Capps	5	Accepted	Including two reported in 1944 paper
1952	Monkhouse	1	Accepted (Capps 1952)	Unpublished Referred to by Capps (1952)
1952	Hallpike	1	Accepted (Capps 1952)	Unpublished Referred to by Capps (1952)
1952	Simpson J F	1	Accepted (Capps 1952)	Referred to by Capps (1952) Later published by Ballantyne J C (1953)
195	Munro Black	6	Accepted	
1952	Rosenwasser	7	Accepted	Also reviewed cases of K B Brown (1951) Hoople (1951) Walsh (1951) and Miller (1951)
1952	Rosen	1	Accepted	Also referred to two others to be published by Israeli Authorities
1952	Brown L A	6	Accepted	
1953	Amman	1	Accepted	
1953	Henson Crawford and Cavanagh	6	Accepted	
1953	Barton and Thee	3	Accepted	
1953	Bickerstaff and Howell	3	Accepted	
1953	Salkeld	1	Accepted	
1954	Hierons	1	Accepted	
1954	Towson and McNeilis	1	Accepted	
1954	Pérez Toledo and Taveras	1	Accepted	
1954	Bradley and Maxwell	10	Accepted	
1954	Strong	1	Accepted	
1954	Gross	1	Accepted	[illegible]
1954	Figi and Weisman	22	Accepted	
1955	Williams Childs Parkhill and Pugh	11	Accepted	
1955	McNeill and Milner	1	Accepted	
1955	Davol	1	Accepted	
1955	Huppler McBean and Parkhill	1	Accepted	
1955	Holesh	3	Accepted	Case 4 proved to be a chordoma (personal communication to authors from Dr S P Meadows)
1955	Rao	2	Accepted	
1956	Stewart Ogilvie and Sammon	7	Accepted	
1956	Conley	1	Accepted	
1958	Simpson I C and Dallachy	3	Accepted	

number of these were accepted as probable or definite examples of glomus jugulare tumour (Table I) The largest single series contained 22 cases and was reported by Figi and Weisman (1954) from the Mayo Clinic It is interesting to note that only three of these 22 cases had originally been diagnosed histologically as tumours of the glomus jugulare The number of cases recorded in the period 1945 56 is 158 (Table II), which with 14 accepted examples reported before 1945 makes a total of 172 Tables I and II are summarized in Table III

Various aspects of the subject have been reviewed by Winship *et al* (1948), Bartels (1949), Lattes and Waltner (1949), Berg (1950) and Gaffney (1953) The largest number of cases considered in these reviews was 56

TABLE III

TUMOURS OF THE GLOMUS JUGULARE IN THE VICINITY OF THE MIDDLE EAR, SHOWING THE NUMBER OF CASES IN THE LITERATURE INCLUDING THE THREE REPORTED IN THE PRESENT PAPER. THIS TABLE SUMMARIZES THE FACTS RECORDED IN TABLES I AND II.

	Accepted cases	Probable cases	Cases not proven
Number of cases of *tumours of the glomus* jugulare reported before Rosenwasser (1945)	14	19	13
Number of cases of tumours of the glomus jugulare reported during and after 1945 (until the end of 1956)	158	10	1
Totals	172	29	14

in the article by Gaffney. Table IV shows the numbers of accepted and probable cases collected from the literature by the authors of these reviews.

Nomenclature. Lattes and Waltner (1949) noted the similarity of the structure of the glomus jugulare, glomus caroticum and glomus aorticum, and pointed out that similar tissue occurred in the ganglion nodosum of the vagus nerve and in the vicinity of the ciliary ganglion. They regarded all these structures as homologous parts of a system of non-chromaffin tissue which was closely associated with the parasympathetic ganglia of the oculomotor, glossopharyngeal and vagus nerves. They suggested that a tumour of these tissues should be called a "non-chromaffin paraganglioma". This term has not been generally adopted. Willis (1953) states that the results of the chromaffin reaction have been variable not only in different growths of similar microscopic structure but in different parts of the same growth. The paraganglia are collections of cells similar to those of the adrenal medulla in development, structure, innervation and

TABLE IV

NUMBERS OF CASES CONSIDERED IN VARIOUS REVIEWS OF TUMOURS OF THE GLOMUS JUGULARE IN THE MIDDLE EAR. A REVIEW OF THE CLINICAL FEATURES OF THESE TUMOURS BY BICKERSTAFF AND HOWELL (1953) WAS BASED ON 90 CASES.

Authors of reviews	Number of acceptable cases reviewed by authors
Winship, Klopp and Jenkins (1948) .	11*
Bartels (1949)	18
Lattes and Waltner (1949)	18
Berg (1950)	17
Gaffney (1953)	56
Present authors	172

* Other 13 probable cases are mentioned; review is admitted by the original authors to be incomplete.

function (Hollingshead, 1940). Such cells occur in the interadrenal sympathetic nerve cells surrounding the commencement of the inferior mesenteric artery, in the enterochromaffin cells of the intestinal mucosa, and occasionally in thoracic sympathetic ganglia. A tumour of these tissues alone may properly be called a "paraganglioma". A tumour of the adrenal medulla is, however, generally termed a phæochromocytoma and one arising from the enterochromaffin cells in the intestinal tract (including the vermiform appendix) is termed an argentaffin carcinoma (or carcinoid). All these tumours show a definite positive chromaffin reaction. Most also elaborate a pressor substance, 5-hydroxytryptamine (Serotonin) which may cause paroxysmal or sustained hypertension. The majority are simple and grow slowly.

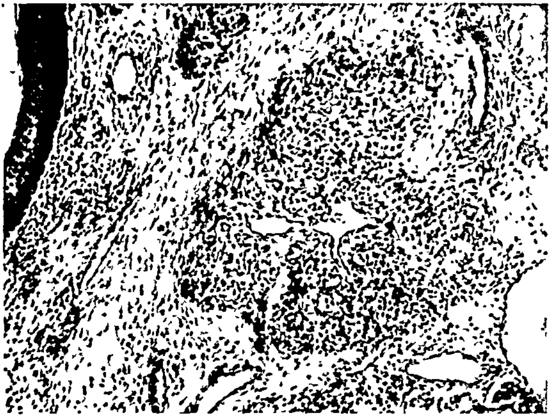

FIG 1

Case 1 Section of the polypoid tumour removed from the left external canal at mastoidectomy in 1937 The covering of stratified squamous epithelium is seen at the top left A large group of loosely arranged "epithelioid" cells, with three sinusoidal blood spaces, is in the central portion Smaller groups of "epithelioid" cells and several large blood spaces occur elsewhere (H and E × 110)

In an attempt to give tumours of chemoreceptor organs a definite name, Mulligan (1950) suggested the term "chemodectoma". This generic term simply means a tumour occurring in a chemoreceptor organ and gives no indication of structure or behaviour. Gaffney (1953) proposed that "receptoma" should be used, and that this generic name should be qualified by the structure from which it arose, e.g. the neoplasm which is the subject of this paper, would be called a "jugular receptoma". However, none of these has been widely adopted and in the British literature the terms "tumour of the glomus jugulare" or "tympanic body tumour" are generally used (Black, 1952; Capps, 1952, Magarey, 1952, Stewart *et al.*, 1956).

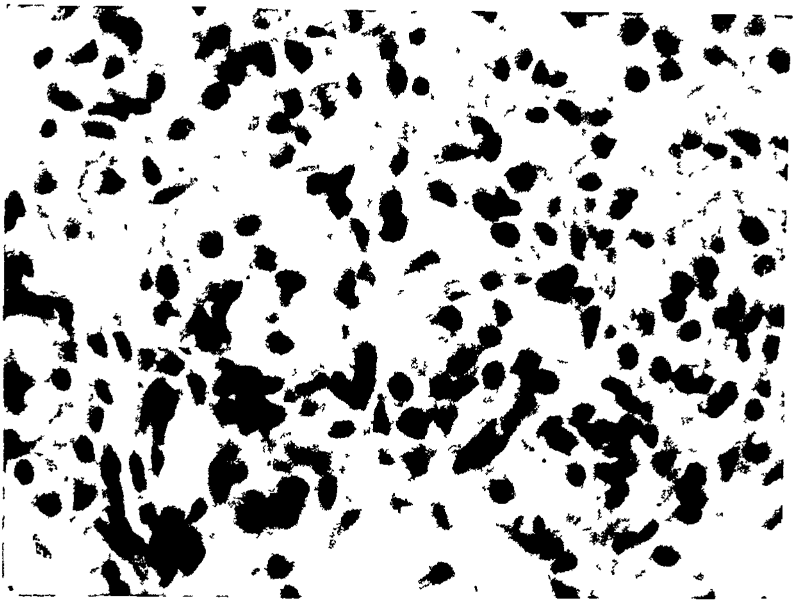

FIG 2

Case 1. Section of tumour removed in 1937 "Epithelioid" cells are fairly large and are polygonal Cytoplasm is abundant Nuclei are rounded or ovoid, most are dark and uniformly stained, a few show granular chromatin No mitoses are present (H and E × 500)

Structure and Histological Diagnosis

The neoplasm is composed of cords or solid masses of "epithelioid" chemoreceptor cells. An alveolar arrangement is fairly common. The solid masses may be round or of irregular outline. The peripheral "epithelioid" cells in the groups are in contact with the endothelium of vascular spaces or poorly supported blood vessels similar in size to capillaries or sinusoids. In many of the reported tumours, these vascular elements were large and conspicuous, and as a result, a diagnosis of hæmangioma was sometimes made. The number and size of the blood vessels vary, not only between different tumours but in the same tumour at different stages of its clinical course, as may be seen in Case 1 of the present paper. The first biopsy from this case was obtained in 1937 and a section is shown in Fig. 1. A second biopsy was taken in 1944, two courses of radiotherapy having been given during the intervening seven years. A section from this second specimen (Fig. 2) shows more "epithelioid" cells and fewer blood vessels

The "epithelioid" cells are large or of moderate size and are rounded or polygonal The cytoplasm is acidophilic and abundant, it is usually clear and uniform but is occasionally finely granular or vacuolated The nuclei are small and vesicular, often with distinct nucleoli, and mitoses are absent The cell boundaries are usually distinct but in most reported tumours there are syncitial areas Using the Masson trichrome stain or one of its modifications, a scanty collagenous stroma is seen between groups of "epithelioid" cells, this stroma does not surround every neoplastic cell (Fig 3) Silver impregnation reveals an argentaffin network of delicate reticulin fibres, precursors of collagen, branching from the subendothelial membranes of the blood vessels and surrounding groups of tumour cells Like the more mature collagen fibres the reticulin fibres do not surround individual tumour cells (Fig 4)

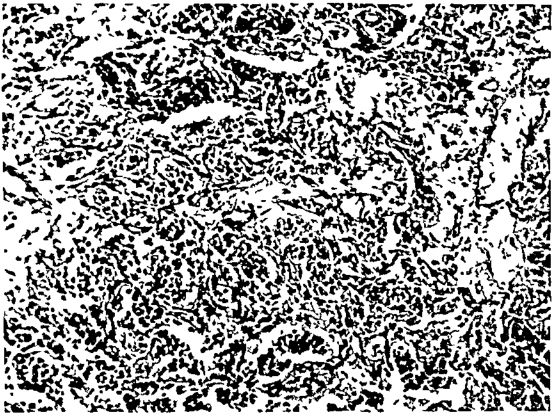

FIG 3

Case 1 Section of polypoid tumour removed in 1944 from the left radical mastoid cavity Epithelioid tumour cells are more numerous and more diffusely arranged than in the sections obtained in 1937 Blood vessels are still numerous (Trichrome × 150)

In tumours which contain vascular granulation tissue silver impregnation demonstrates the boundary between the granulation and neoplastic tissue A few lymphocytes are usually present throughout the substance of these tumours and fibroblasts and elastic fibres are present in small numbers Neither fat nor glycogen has been demonstrated

The so-called chromaffin reaction of tumours of the glomus jugulare has been investigated in only a few cases. The reaction refers to the yellowish-brown colour assumed by certain cells and tissues after being fixed in fluids containing chrome salts. It is a specific chemical test for polyphenols, aminophenols and *o*- and *p*-polyamines. Chromaffin granules cannot be demonstrated by post-chroming sections or blocks which have been fixed in fluids not containing chrome salts (Pearse, 1953). The specimens from the three cases described in this paper were received in formalin and the chromaffin reaction was therefore not investigated since the results would have been invalid. The reaction applied to chemoreceptor tissue has been studied more often in the normal carotid body and in tumours of the carotid body in which relatively large amounts of material are available for investigation. The result of the reaction in normal carotid bodies is variable, differs in different animals, but is never strongly positive (Le Compte, 1948). Only three substances in the body give a true positive reaction:

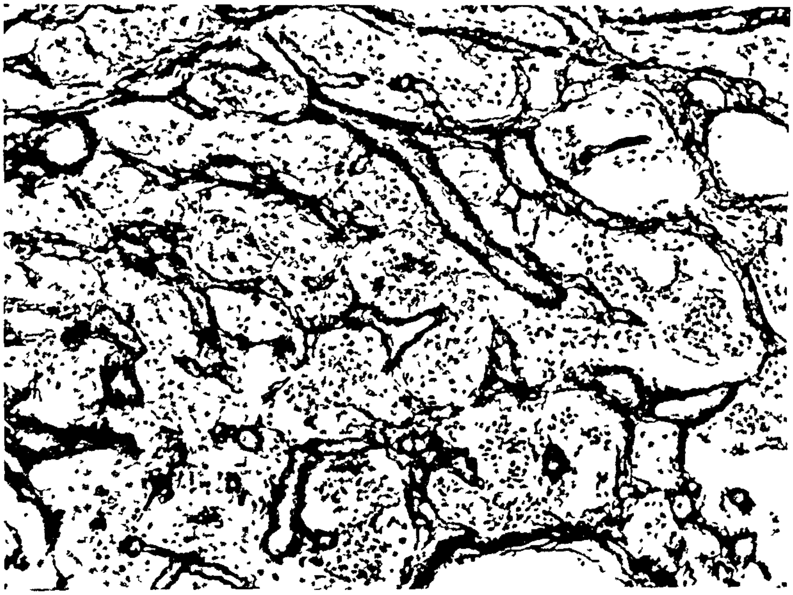

FIG 4

Case 1 Same general field as Fig 3 showing the very vascular pattern outlined by the reticulin fibrils, no reticulin fibrils are seen between individual "epithelioid" tumour cells (Laidlaw's reticulin stain × 150)

(i) the adrenalin precursor substances in the adrenal medulla;
(ii) the adrenalin precursor substances in the paraganglia; and
(iii) the cytoplasm of the specialized granular cells (enterochromaffin cells) found in the crypts of Lieberkühn in the small intestine.

Metastases and Coincident Tumours

Tumours of the glomus jugulare in the middle ear grow slowly and the constituent cells are uniform in appearance There is no capsule, however, and continuous expansion of the growth occurs with extension outwards along the external auditory canal, upwards into the middle cranial fossa, medially with damage to or destruction of the structures of the internal ear and labyrinth, or downwards into the jugular foramen Spread may occur in more than one direction and further details are given in the section on the "Clinical Course"

The neoplasm, in the vast majority of recorded cases, shows locally invasive characteristics The case reported by Seiffert (1934) showed infiltration of the wall of the jugular vein with accompanying thrombosis and it is theoretically possible for neoplastic cells which invade vessel walls to break off into the blood and to be disseminated in this way There is considerable doubt whether neoplastic cells from glomus jugulare tumours do, in fact, grow in the organs to which they are presumably carried by the blood stream There are many examples of extensive spread of the original tumour within the cranium but in only four instances have metastases been reported

These four cases in which metastatic growths have been recorded are listed in Table V In the case described by Winship *et al* (1948), the photomicrograph of a section from an enlarged cervical lymph node shows histological appearances consistent with the diagnosis of glomus jugulare tumour Unfortunately no biopsy was taken from the tumour of the middle ear which was treated by radium needle implants in the external auditory canal The photomicrograph of the metastasis in the liver in Case 3 of Lattes and Waltner (1949) is not convincing but it is possible

TABLE V

Cases of Glomus Jugulare Tumours in which Metastases have been Reported

Authors	Site of origin	Site of metastases	Remarks
Winship Klopp and Jenkins (1948) Case 1	L aural polyp	L jugular lymph nodes (biopsy of one node)	No biopsy of aural tumour
Lattes and Waltner (1949) Case 3	L aural polyp	One large metastasis in liver	Photomicrograph of metastasis is not convincing
Tamari McMahon and Bergendahl (1951) Case 1	R aural polyp	Microscopic metastases in liver and lungs	No photomicrographs
Henson Crawford and Cavanagh (1953) Case 1	L aural polyp	Liver lungs and spleen	No histological examination of metastases

that the atypical microscopic appearances are due to the failure of continuing autonomous growth of a focus of glomus jugulare tumour cells and that the cells are degenerating; another possibility, which is mentioned by Gaffney (1953), is that the original aural polyp is an extension of an intracranial carcinoma. The case recorded by Tamari *et al.* (1951) is not accompanied by photomicrographs of the metastatic deposits and the description of these is brief and occurs in the general discussion of the case. In Case 1 described by Henson *et al.* (1953) there was no microscopic examination of the metastases; the histological appearances of the tumour of the left glomus jugulare and of its intracranial extension are illustrated and are acceptable.

It is concluded from this study of the literature that tumours of the glomus jugulare grow slowly, are locally invasive and may extend for a considerable distance from the site of origin. There is, however, no conclusive evidence that they give rise to distant metastasis.

Coincident Tumours. Several cases have been reported in which a tumour of the glomus jugulare in the middle ear was associated with a tumour in one of the other chemoreceptor organs.

Lubbers (1937) described a patient who had a tumour of the carotid body on one side and a tumour of the middle ear on the opposite side. The histological structure of the two tumours was identical. Lubbers thought that the carotid body tumour was one of the rare malignant examples and that the growth in the middle ear was a metastasis. This seems to have been the first occasion on which a resemblance was noted between a vascular tumour of the middle ear and a carotid body tumour. The clinical aspects and pathological material in this case were re-examined by Bartels (1949) who decided that the growth in the middle ear was a tumour of the glomus jugulare which had occurred in a patient who, by chance, also had a carotid body tumour. Bartels included this case in his own series.

Another patient who had a glomus jugulare tumour of the middle ear with a swelling in the region of the carotid body of the opposite side is mentioned by Capps (1952). This patient was in the care of J. F. Simpson and details of the case have not been published. Kipkie (1947) reported the simultaneous occurrence of tumours of the carotid body and of the glomus jugulare in one patient and Lattes and Waltner (1949) described a similar case.

Conley (1956) described a female aged 26 who had a simple tumour of the left carotid body which had been present for six months; it was completely and successfully removed. Eight months after the operation a glomus jugulare tumour arising from the dome of the jugular bulb in the floor of the right tympanic cavity was discovered and removed without untoward effects.

Bilateral tumours of the glomus jugulare in the middle ear have been reported twice One was described by Bartels (1949) and the other by McNeill and Milner (1955) In the latter, biopsy of one of the tumours was followed by profuse hæmorrhage Since similar hæmorrhage might reasonably have been expected from the second tumour, biopsy of it was not performed

Multiple tumours of paraganglionic tissues which, however, did not involve jugular glomera have been reported in two patients by Lattes (1950) One of these had a tumour of the ganglion nodosum of the right vagus and a carotid body tumour on the opposite side The other patient had three separate tumours—one arising in the region of the ganglion nodosum of the vagus, one at the carotid bifurcation and the third in the adventitia of the aortic arch Lattes *et al* (1954) recorded the details of a patient who had a carotid body tumour in the neck and a tumour of similar microscopic structure in the orbital fat

Marcuse and Chamberlin (1956) reported a case which had bilateral carotid body tumours and three separate nodules with similar histological structure, one of these was attached to the left vagus 50 mm inferior to the left carotid bifurcation, and the other two were close to the upper pole of the left carotid body tumour These authors found ten previously recorded cases of bilateral carotid body tumours in the literature

Single tumours of chemoreceptor tissue have been reported arising in other sites in the ganglion nodosum of the vagus nerve at the jugular foramen (Lattes 1950, Stewart *et al*, 1956), in the aortic glomera in the mediastinum (Lattes, 1950, Duncan and McDonald, 1951), in the ciliary body in the orbital fat (Fischer and Hazard, 1952), in the retroperitoneal fibromuscular tissues of the pelvic wall (Block *et al* 1955) and in the chest wall (Shaw and Kennedy, 1956) Smetana and Scott (1951) and Randall and Walter (1954) have reported tumours of similar structure arising in the muscles of the thigh and neck, some of which gave rise to distant metastases, unlike paragangliomas arising from other sites A single doubtful case has been reported in the larynx (Andrews 1955)

Clinical Course

These tumours occur mainly in the middle aged—22 69 years according to Lundgren (1949) and 45-50 according to Towson and McNeilis (1954) Women are affected five times more commonly than men

They vary in their rate of growth but the development of the clinical features follows a general pattern, individual variations being due to differences in location of the jugular body affected They are usually slow in development and extension is by direct local expansion

Deafness of conductive type and tinnitus are the earliest symptoms and they may remain the only abnormalities for many years A sensation of fullness and of pulsation in the ear has been noted but pain either local or referred, is uncommon, although Capps (1952) found it in early cases. In the early stages the drum appearance is normal and it remains so until the tumour is large enough to shine through as a red discoloration. Before the appearance of an actual tumour the drum has been described as presenting a gunmetal blue coloration (Magarey, 1952). As the neoplasm develops, it gradually presents as a definite dark red intratympanic tumour with the drumhead stretched over it. Usually the mass is seen behind the lower part of the drum as the jugular bulb glomus is the commonest site of origin. Myringotomy has been performed at this stage in the mistaken diagnosis of an acute inflammatory process. The drum picture may remain unchanged for 12-18 months (Magarey, 1952). At this stage the drumhead may be lifted off the tumour by the use of the Siegle's speculum and the "pulsation sign" described by Brown (1953) is sometimes noted.

Destruction of the overlying drum follows by direct pressure and the tumour itself presents in the external canal. Bleeding may occur at this stage either spontaneously or on local trauma. Infection of the middle-ear cavity usually occurs later when the appearances are commonly mistaken for those of a chronic suppurative otitis media with aural polyp formation or even for carcinoma, the discharge being frequently blood-stained. The removal of the supposed polyp or surgical exploration of the mastoid is usually accompanied by profuse hæmorrhage from the highly vascular tumour. The case may present as an acute flare-up in a chronic suppurative otitis media and may develop any of the classical complications of the infective process quite apart from the presence of the tumour.

While the neoplasm is growing outwards and visibly, it may extend on its deep aspect destroying the middle ear structures and the labyrinth capsule, and spread slowly along lines of least resistance throughout the petrous pyramid. The VIIth nerve is frequently involved by direct pressure. Growth may advance down the jugular foramen and along the base of skull and extensive intracranial spread may occur. This deep extension may develop early in the history of the case or may appear only in the very late stages. One case has been described in which the tumour grew down the eustachian tube and presented in the nasopharynx (Weille and Lane, 1951).

While the site of origin largely determines the direction in which the growth spreads, the cases fall into four main clinical groups, according to Bickerstaff and Howell (1953), who analysed 90 cases from the literature.

Group I. Aural symptoms only (49 cases).

Group II. Neurological involvement developing many years after aural symptoms (20 cases). The nerves involved by pressure were usually the IXth, Xth, XIth and XIIth.

Sometimes there is, in addition, a swelling in the neck behind the angle of the jaw associated with pulsation and bruit (The VIIth nerve was not included by these authors who considered facial paresis to be an expression of the local middle-ear involvement)

Group III Neurological symptoms and aural symptoms developing concurrently (17 cases) The growth in this group probably arises near the origin of the tympanic nerve

Group IV Neurological symptoms appearing before aural symptoms This group is the least common and only four examples had been reported when Bickerstaff and Howell published their paper in 1953 Since then, Hierons (1954) reported a case presenting with papillœdema and obscuration of vision That author knew of two other similar cases

The onset of deep involvement is heralded by the appearance of cranial nerve palsies and possibly vertigo which may, in some cases, be very troublesome and is due either to direct involvement of the labyrinth or VIIIth nerve

The VIIth nerve, as would be expected from its close relationship to the site of origin of these tumours, is most frequently involved A facial palsy is frequently reported following operation probably due to damage to a nerve already exposed by the advancing tumour Fifth and VIth nerve involvement is more rarely reported and may be caused either by the deep petrous extension or to an intracranial break through with increased pressure The IXth, Xth and XIth nerves are very commonly involved and any combination of partial or complete palsies of one or all of them may be found These nerves are usually affected by direct pressure in the jugular foramen Huppler *et al* (1955) described a unique case which presented as a unilateral vocal cord paralysis Paralysis of the XIIth nerve has been reported from spread along the base of skull (Lattes and Waltner, 1949, and Williams *et al*, 1955) In one case (Figi and Weisman, 1955) all cranial nerves from the IIIrd to XIIth were involved

Although the symptomatology is non specific four characteristic features of these tumours were emphasized by Lattes and Waltner

(1) An extreme tendency to bleed following the slightest trauma
(2) A subjective sense of pounding and pulsation in the ear
(3) A progressive, expanding growth leading to destruction of the temporal bone through pressure atrophy and eventual intracranial extension with compression of central nervous structures and cranial nerves

(4) A usually lengthy duration of symptoms frequently associated with recurrences of polypoid masses in the external canal following repeated removals.

In some cases the deep extension with secondary nerve palsies may be much greater than the spread outwards and extensive petrous destruction may be present with little change in the drum appearances.

Radiographic examination, which should always be carried out, may show the bone destruction in the petrous pyramid and jugular foramen.

Differential Diagnosis

The clinical features described above follow the general course but too much weight should not be placed on individual signs—the diagnosis must rest finally on histological findings.

The early drum appearances are suggestive but an intratympanic tumour pushing forward the drumhead is not necessarily of glomus origin. Marked bleeding after removal of a supposed polyp in the more advanced case calls for an immediate biopsy, but hæmorrhage is not invariably found as instanced by two cases of Williams *et al.* (1955) and our own Case 3.

Cranial nerve involvement is not, of course, of diagnostic significance in itself. It must be remembered that these cases may be seen at any stage of their development and the picture may be markedly obscured and complicated by superadded middle-ear and mastoid infection, while on the other hand, an isolated cranial nerve palsy may be the immediate presenting feature.

Conditions which may simulate glomus jugulare tumours include: chronic suppurative otitis media with aural polyp formation; an acute inflammatory process; carcinoma of the middle ear or external canal; VIIIth nerve tumours and tumours of the cerebellopontine angle; carcinoma of the nasopharynx; basal meningioma; metastases in the basal meninges; syphilitic meningitis; neurinoma of IXth, Xth, XIth, or XIIth nerves; carcinoma of the sphenoid sinus; chordoma; Hand-Schüller-Christian disease; and Ewing's tumour (two cases reported in the mastoid process by Geschickter and Copeland, 1949).

Treatment

There has been considerable variance of opinion on this, the essential question lying in the radiosensitivity of the tumour. The histological appearances would suggest that the tumour is radioresistant; it has for long been so considered and indeed many radiotherapists still regard irradiation as of little value. On the clinical side, however, there has been a gradual move away from the belief in surgery alone in the deeply extending advanced growths.

An attempt at complete excision was advocated by Lundgren (1949), Weille and Lane (1951) and Winship and Louzan (1951). Rosenwasser (1952), Barton and Thee (1953) and Bradley and Maxwell (1954) found that radiotherapy produced no demonstrable effect but Graf (1950) reported a favourable response in eight cases. Poppen and Riemenschneider (1951), although doubtful of its value, advised radiotherapy in more advanced cases.

Capps (1952) stressed the high mortality of extensive operative procedures and considered that radiotherapy alone should be relied upon in extensive disease; surgery and therapy should be combined for the earlier cases, with further irradiation for recurrences.

The previous insistence on complete surgical extirpation appears to be modifying and the present position lies between the two extremes. In early cases with limited, accessible growths, radical mastoidectomy with removal of tumour, followed by deep therapy would appear to be the treatment of choice. X-ray (or radium) may be omitted, however, in the first instance but used on any sign of recurrence.

In the more advanced case (without extensive deep erosion as demonstrated radiographically) as a preliminary to deep therapy, radical mastoidectomy with removal of as much tumour as possible may be carried out with avoidance of heroic attacks on the petrous temporal bone and the jugular bulb. Weille and Lane (1951) emphasized the great danger to the patient of extensive procedures around the bulb. The main hazard at operation is the profuse bleeding, and preliminary carotid ligation and even lateral sinus occlusion have been carried out with little effect. Stewart *et al.* (1956) suggest that deep therapy prior to surgery might permit a less hazardous operation by diminishing the blood supply. These authors also stress the danger of extensive surgical "adventures" on advanced growths and point out the age factor and slow growth of the tumour as factors which must be weighed carefully in considering treatment.

In cases showing advanced deep extension, surgery is likely to be of little value and fraught with great immediate danger to life and a trial of deep therapy alone is the treatment of choice.

According to Williams *et al.* (1955), a dose of 1,200 r to 2,400 r is considered adequate for these tumours.

In considering the question of the effect of irradiation on these growths, Figi and Weisman (1954) emphasize that radiosensitivity should not be confused with "radiocurability". It is unlikely that any case was "cured" by therapy alone but there seems ample clinical evidence that the rate of growth is slowed and recurrences kept under some degree of control. It may be that this clinical response is due not to any radiosensitivity of the active tumour cells but to a diminution in the vascular supply following irradiation.

I. C. Simpson and Russell Dallachy

Case Reports

CASE 1

Female, aged 37, when first seen because of right otorrhœa and deafness for four years associated with a right aural polyp. Removal of "polyp" and a radical mastoidectomy were accompanied by severe hæmorrhage, and followed by purulent meningitis which responded to chemotherapy. Polyp in mastoid cavity recurred after seven years and removal was again accompanied by severe hæmorrhage. Two courses of radiotherapy caused regression of the tumour for a further nine years, but the tumour again recurred. Duration of follow-up: 19 years.

Mrs. M.A., aged 37 years when first referred to the Victoria Infirmary on 10.11.37 complained of constant right otorrhœa of four years' duration; there was also marked deafness and tinnitus but there had never been either pain or vertigo. Examination revealed that the right external auditory canal was obstructed by a large "aural polyp" and a diagnosis of chronic suppurative otitis media with a simple pyogenic polyp was made.

First operation. On 11.11.37 a postaural radical mastoidectomy was carried out and bleeding was profuse throughout the operation. The pathological diagnosis of the polyp and tissue from the mastoid was "fibroangioma".

Six hours after the operation, a right facial palsy developed. On the following day (12.11.37) the patient complained of vertigo, and nystagmus was noted. On 17.11.37 vomiting and pyrexia were added to the above features and on 20.11.37 lumbar puncture revealed a turbid cerebrospinal fluid with a cell count of 3,300 per c.mm. (92 per cent. polymorphs). No organisms were obtained on culture. The meningitis was treated with "Prontosil" and recovery was rapid.

The patient was not seen again for seven years but on 24.10.44 she reported because of further polyp formation in the radical cavity.

Second operation. On 8.11.44 under general anæsthesia, the polypoidal tissue was removed by Mr. T. O. Howie. Bleeding was severe. The pathological diagnosis of the tissue removed was "hæmangioendothelioma".

Post-operative treatment and progress. During December, 1944, a course of radiotherapy was given, using an anterior and posterior field, 1,500 r tumour dose in each field over three weeks. The tumour regressed markedly. In March, 1945, further therapy was given using the same fields, 1,000 r to each field over two weeks.

The radical cavity became dry and only a small "angiomatous polyp" was present in the attic. The patient, from this time, was kept under regular supervision and the small remnant of growth never completely disappeared. A tendency to hæmorrhage was repeatedly noted on any manipulation in the ear. The tumour showed no enlargement until February, 1954, when the whole canal was found filled with a large polypoid mass apparently covered by epithelium. No discharge from the cavity was noted. In October, 1954, a further course of deep X-ray therapy was given using a single field, 1,500 r tumour dose over two weeks, with only slight regression in the size of the tumour.

In February, 1955, an acute aural infection developed and an abscess in the lower part of the postaural wound had to be drained. Apart from this, no further complications have arisen and the tumour which fills the external canal remains unchanged.

The clinical re-assessment of this case as a possible example of a glomus jugulare tumour prompted a complete review of all the pathological material from the original blocks and sections. A fresh biopsy was not advised because of the danger of hæmorrhage.

Revised pathological report on "polyp and tissue from mastoid" (removed at operation in 1937). The microscopic appearances are those of a tumour of the glomus jugulare. The neoplastic cells occur in loosely arranged clusters of varying size; there is no arrangement into cords or alveoli (Fig. 1). Blood vessels are present in large numbers throughout; most are capillaries but a few sinusoidal vessels occur. Between the masses of neoplastic cells there is plentiful loose fibrous stroma which is fairly cellular and contains a moderate number of round cells and also erythrocytes which have come from the numerous vessels The neoplastic cells are all of moderate size, have a delicate cell membrane which is not readily seen and have finely granular, acidophil cytoplasm. Most of the nuclei are rounded, and densely and uniformly stained; a small number are ovoid or elongated with granular chromatin. Mitotic figures do not occur (Fig. 2).

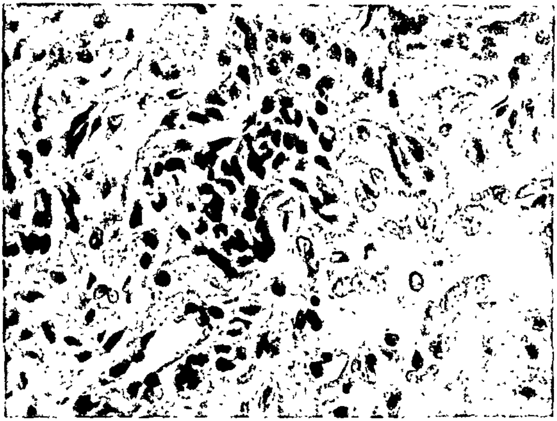

FIG. 5

Case 1. Section of tumour removed in 1944. "Epithelioid" tumour cells have nuclei which are more vesicular than in the tumour removed in 1937. The cells in the centre of the field with pyknotic nuclei are similar to those seen in the previous specimen (H. and E. × 500).

Revised pathological report on "polyp" removed in 1944. Microscopic examination shows the neoplasm to be more cellular than the tissue obtained in 1937. The neoplastic cell masses are larger and are not so well demarcated.

The fibrous stroma is denser and less cellular (Fig. 3). The nuclear chromatin of the neoplastic cells is more granular and nuclei are paler (Fig. 5). Silver impregnation demonstrates the reticulovascular meshwork throughout the neoplasm. The argentaffin reticulum fibres are seen surrounding fairly large groups of neoplastic cells and neither these fibres nor those of mature collagen are present between individual tumour cells (Fig. 4).

CASE 2

Female, aged 42 when first seen because of increasing deafness in the right ear for four years associated with a right aural polyp. Removal of polyp was accompanied by profuse hæmorrhage. One week later, an acute mastoiditis with a Von Bezold abscess had developed and a postauricular radical mastoidectomy was carried out. Two weeks after the second operation, exuberant "granulations" and the remains of the "polyp" were removed from the mastoid cavity. A pathological diagnosis of "well differentiated adenocarcinoma" was made and a single course of radiotherapy was given. The radical cavity epithelialized completely and there was no recurrence of the tumour. Duration of follow-up: 17 years.

Mrs. A.S., aged 42 years when admitted to the Victoria Infirmary on 24.4.40, gave a history of deafness in the right ear which had been increasing for four years. Four weeks before admission, the ear had been syringed and had bled for several days afterwards: an "aural polyp" was diagnosed and its removal was advised. Two weeks before admission, watery discharge from the ear was reported for the first time and some improvement in hearing was noted. During the week before admission a tender swelling appeared in the right side of the neck.

Condition on admission. A large "aural polyp" filled the right external auditory canal; the left ear was normal. Nose, pharynx and nasopharynx showed no abnormality. A large, tender, fluctuant mass was present in the right side of the neck. Radiography revealed a densely sclerotic mastoid on the right side. Total white cell count was 21,000 per c.mm.

First operation. On 25.4.40, the "polyp" was removed. The operation was accompanied by profuse bleeding. The pathological diagnosis of the material submitted for examination was "cellular hæmangioma modified by infection".

During the week following operation, the patient developed a high pyrexia and her general condition deteriorated. She was seen by Mr. T. O. Howie who advised immediate exploration of the right mastoid.

Second operation. On 1.5.40, a right postauricular radical mastoidectomy was performed (Mr. T. O. Howie). Considerable bone necrosis and large masses of "granulation tissue" filling the middle ear, attic and antrum were reported. A Von Bezold abscess was found and drained through a counter incision in the neck. The postaural wound was packed open and the remains of the "polyp" in the external canal were left undisturbed.

The patient's general condition immediately improved. A delayed right facial paresis developed but this was transient. On 16.5.40, further "granulations" and the remains of the "polyp" were removed from the middle ear and mastoid cavity and a secondary suture of the wound carried out and thereafter convalescence was uneventful. The pathological report on the tissue removed from the mastoid cavity stated that the appearances were strongly suggestive of a fairly well-differentiated adenocarcinoma with numerous solid acini set in granulation tissue

Post operative treatment and progress In view of the pathological report the patient underwent a course of radiotherapy, 4,300 r being given by a single field The radical cavity epithelialized completely When the patient was seen in May, 1957, the radical cavity was still dry and free from tumour growth and there was no radiographic abnormality of the petrous region It was considered that this patient might have had a glomus jugulare tumour and

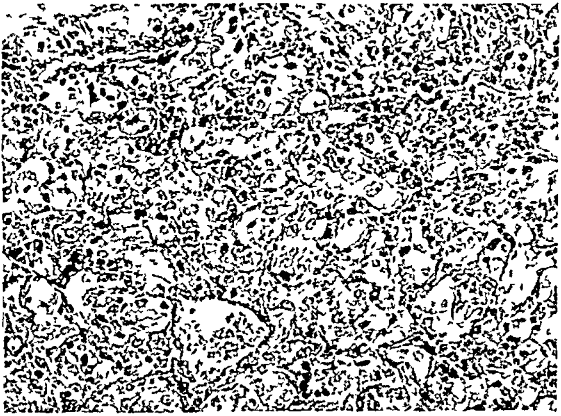

Fig 6

Case 2 Section of glomus jugulare tumour removed from right mastoid cavity in 1940 The epithelioid [illegible] i those in Case 1 and grouping of cells i [illegible] blood vessel is seen at the bottom t [illegible] picro Mallory × 250)

so fresh sections were obtained from the original blocks of tissue removed in 1940

Pathology The microscopic appearances of the *polyp removed from the right external auditory canal* (on 25 4 40) are those of an infected vascular polyp in which there are a few small collections of cells which resemble the ' epithelioid cells of a glomus jugulare tumour The lesion consists mainly of loose and œdematous connective tissue in which there are numerous poly morphs plasma cells and macrophages Many cleft like capillaries are also present A narrow zone of condensed fibrous tissue surrounds some of these vessels Close to the endothelium there are a few small groups of cells which are loosely arranged. The cells are similar in size to macrophages, and are ovoid; they have uniformly staining eosinophil cytoplasm and small, rounded nuclei which are dark and appear pyknotic. Reticulin fibrils surround groups of 6-12 cells but do not surround individual cells. The number of these cell groups is small and individual cells in groups are few. These facts together with the pyknotic changes associated with secondary infection do not allow a definite diagnosis of a glomus jugulare tumour to be made. Nevertheless, it is likely that these cells are the "epithelioid" cells of such a tumour especially when the features of the material from the mastoid cavity are considered.

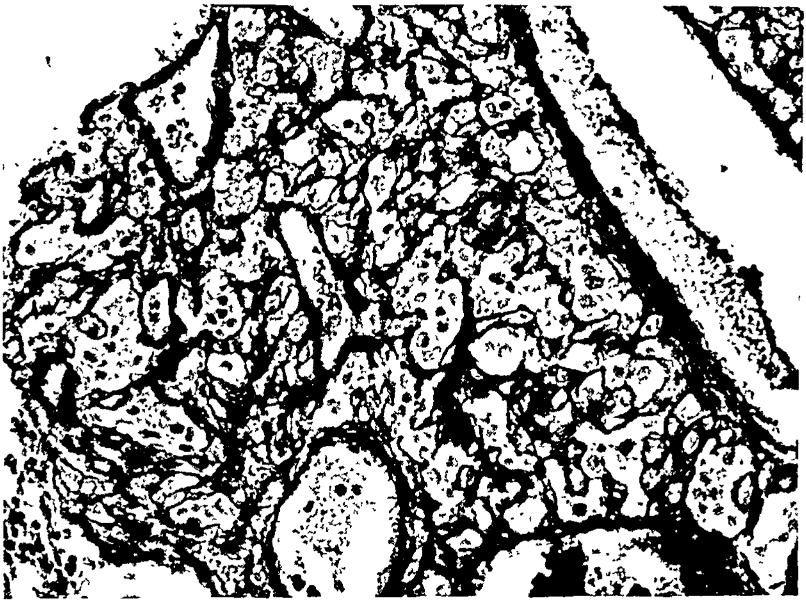

FIG. 7.

Case 2. Section from a field comparable to that shown in Fig. 6. The reticulin network is shown extending from the subendothelial membrane of the blood vessel walls and surrounding groups of "epithelioid" tumour cells but not enclosing individual cells (Laidlaw's reticulin stain × 250).

The histological features of *polypoid material from the mastoid cavity* (16.4.40) are those of a glomus jugulare tumour. The lesion is composed of rather ill-defined rounded groups of cells, each group being separated from its neighbours by only a small amount of vascular collagen (Fig. 6). The cells are rounded or polyhedral, have fairly distinct cell membranes, and uniformly stained or finely granular cytoplasm. The nuclei are of moderate size, are rounded or ovoid, have a distinct nuclear membrane, and have finely dusted, granular chromatin; one or two prominent nucleoli are present, but no mitotic figures are seen (Fig. 8). Reticulin fibres originate from the sub-endothelial zone of the blood vessels and surround groups of 4-10 neoplastic cells but do not surround individual cells (Fig. 7).

CASE 3

Male, aged 63 when first seen because of intermittent bilateral deafness which was diagnosed as eustachian tubal obstruction secondary to maxillary antral infection; both drumheads were normal. One year later, deafness had increased in left ear and a small red tumour was present behind the left tympanic membrane. Removal by endaural attico-antrotomy was not accompanied by troublesome hæmorrhage. Post-operative radiotherapy was given. A reddish area on the drumhead may be a remnant of the tumour but there has been no evidence of further activity. Duration of follow-up: 2 years.

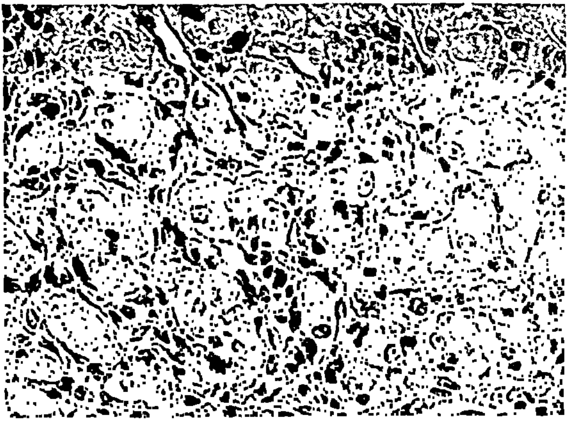

FIG 8

Case 2. Section of tissue removed from right mastoid cavity "Epithelioid" tumour cells are large, polygonal and have fairly distinct cell membranes Cytoplasm is abundant and finely granular Nuclei are rounded or ovoid, vesicular, and there are no mitotic figures (H and E × 500).

Mr. J.C., aged 63 years when first referred to one of us (I.C.S.) at an associated hospital, gave a history of intermittent deafness in both ears for many years During the preceding year deafness had become constant in the left ear and tinnitus developed. Nasal obstruction and discharge had been troublesome for several years. He had no pain, headache or vertigo and no history of otorrhœa.

Examination. The right ear showed a retracted drumhead. In the left ear, the drumhead was rather full posteriorly but otherwise no abnormality was noted. Rinné's test was negative on both sides. Weber's test was lateralized to the left. Nose: mucopus in both airways. Pharynx: slight generalized injection. Post-nasal space and larynx were both clear.

Inflation was not carried out in view of the nasal infection but clear mastoid X-rays and grossly thickened antral mucosa in sinus films supported the clinical diagnosis of tubal obstruction secondary to a sinus infection.

Treatment and progress. April, 1954: symptoms remained unchanged and the left drumhead was still "full". Double antral lavage revealed mucus in the right antrum and frank pus in the left.

May, 1954: symptoms had not been affected by the previous lavage. The drumhead on the left side was now in good position and showed no abnormality.

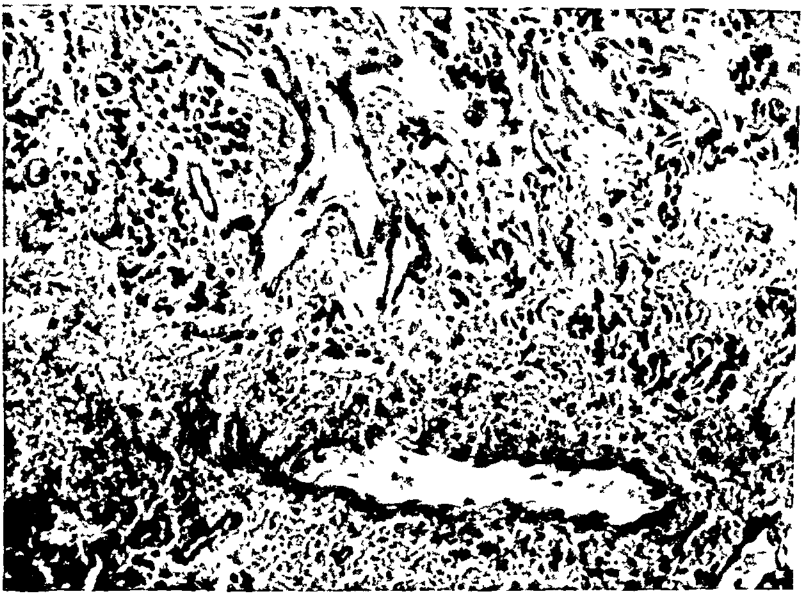

FIG. 9

Case 3 Section of intratympanic tumour removed at endaural attico-antrotomy. The upper half of the field contains groups of "epithelioid" tumour cells in close proximity to numerous capillaries and sinusoidal blood vessels. In the lower part of the field there is a large hæmorrhagic area surrounding a sinusoidal space (Trichrome × 220)

Tuning fork tests remained as on the first examination. A second antral lavage still showed pus in the left antrum.

The patient was asked to report the following month but failed to keep his appointment. He was not seen again until one year later when he was referred in May, 1955, by his own practitioner because the deafness and tinnitus in the left ear had been increasing steadily. No new symptoms had been noted.

On examination (May, 1955) a completely different picture was seen in the left drumhead. A dark red "nodular" tumour was present behind the left tympanic membrane apparently extending downwards from the posterior attic region, the lower drumhead being normal. By use of the Siegle's speculum the drum could be "wrapped around" the mass but could not be lifted from it by negative pressure. As the nose was now clean, left eustachian catheterization was-carried out and the drum position raised—the tumour became much less distinct but a red discoloration shining through clearly marked its presence. The appearances were definitely those of an intratympanic tumour and not of a change in the membrane itself. Radiographs of the mastoid regions still showed no abnormality. A tentative diagnosis of glomus jugulare tumour was made and the patient was admitted to the Victoria Infirmary of Glasgow As the mass appeared to be developing in the upper middle ear and attic it was decided not to attempt biopsy by reflection of the drumhead but to proceed to an endaural exploration

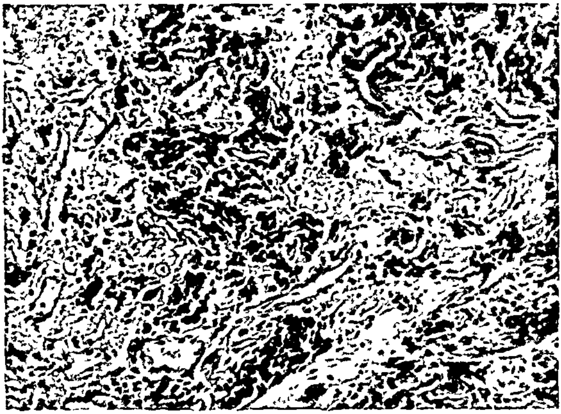

FIG 10

Case 3 Another field from the section shown in Fig 9 Irregular bands of collagen separate the groups of "epithelioid" tumour cells in the right half of the field Areas of hæmorrhage are present elsewhere (Trichrome × 200)

Operation. On 17.6 55, endaural attico-antrotomy was carried out (I C S) The antrum and aditus were healthy The bridge was taken down revealing a small mass of what appeared to be granulation tissue rising from the middle ear behind the drumhead and spreading around the incus The incus was removed to allow access and the portion of tumour in the middle ear was "hooked up" from behind the drum with a seeker. This was achieved without difficulty and all the visible tumour removed as far as could be ascertained The operation was completed in the usual manner—outer attic wall and head of malleus being removed The drumhead was left intact—it appeared normal and no evidence of tumour could be seen through it A remarkable feature in view of the pathological report was the absence of any troublesome hæmorrhage during the removal of the actual tumour.

Pathological Report. The microscopic appearances are those of a neoplasm of chemoreceptor tissue. The lesion consists of numerous capillaries a sinusoidal vascular spaces, most of which are filled with erythrocytes. Th vascular structures are lined with endothelial cells and have a thin layer fine fibrous tissue outside the endothelium. Between the vascular spaces, th are small, loosely arranged masses of "epithelioid" cells (Figs. 9 and 10). Th are polygonal or rounded, and have a moderate amount of clear acidophi cytoplasm. Nuclei are uniformly stained and show no mitotic figures In t more tightly packed cell groups, the cells are smaller and have distinct da ovoid or fusiform nuclei, and have a moderate amount of acidophil cytopla

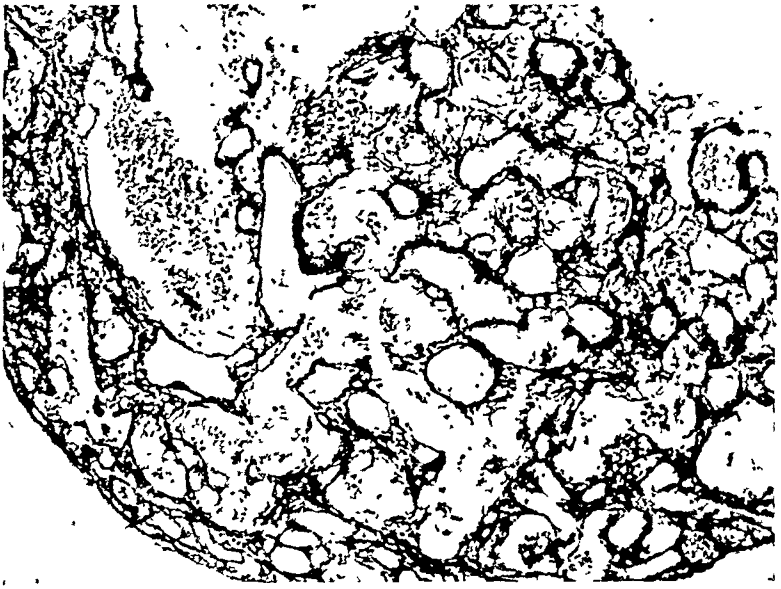

FIG 11

Case 3 Section from a field comparable to that shown in Fig 9 There is considerable hæmorrhage, but the reticulin fibres originate from the subendothelial membranes of the blood vessels and surround groups of "epithelioid" tumour cells (Laidlaw's reticulin stain × 150).

of uniform, non-granular appearance, the cell membranes are not distin (Fig. 12). In the more loosely arranged groups there is slight variation in th size of the cells, and some of the nuclei are granular. Reticulin fibrils from th walls of blood vessels surround groups of neoplastic cells (Fig. 11).

Post-operative treatment and progress. A course of radiotherapy was give using a single small field (2,000 r tumour dose over two weeks). At the tim of writing this paper, the attico-antrotomy cavity is dry and complete epithelialized. A persisting reddish coloration of the upper drumhead suggest a residual portion of tumour may still be present.

This case is reported in detail as few, if any, have been seen at such a early stage when within one year the ear was seen before and after the appearance of an intratympanic tumour.

Review of Tumours of Glomus Jugulare

Summary

Until quite recently, tumours of the glomus jugulare were regarded as rare. This was largely because the histogenesis was not understood and because the histological structure is variable. The literature on the subject has been confusing, as in several of the earlier cases the same tumour has been described in more than one paper, occasionally by different authors and even under different names. In the present work, all the examples in the literature are arranged in the chronological order in which they were first recorded but a comment is made where a case has appeared in more than one paper and where the correct diagnosis is in doubt. Thus the total number of cases cannot be found merely by adding the numbers in column 3 of Tables I and II.

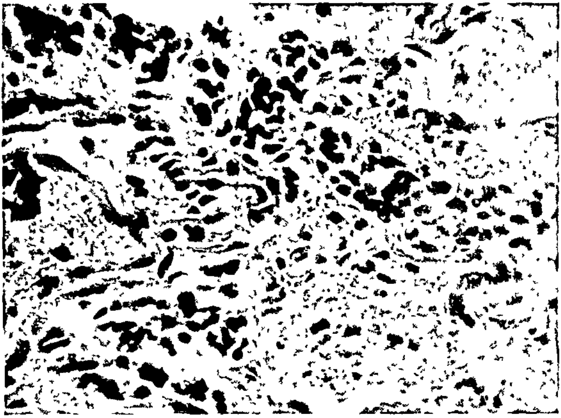

FIG 12

Case 3 Groups of "epithelioid" tumour cells with densely stained nuclei are seen in the upper part of the field to the right of centre A thin-walled blood vessel containing erythrocytes is at top right An endothelial-lined space is in the left upper quadrant Considerable interstitial hæmorrhage is present (Trichrome × 500)

The tumours arise in the glomus-like body situated in the petrous temporal bone in the vicinity of the superior bulb of the jugular vein close to the middle ear. The normal glomus jugulare is similar in its microscopic structure to the carotid body, and its innervation and blood supply are derived from the same sources. The carotid body is a chemo-receptor organ and it is believed that the glomus jugulare has a similar function although a much smaller circulatory region is involved; no hæmodynamic dysfunction has so far been ascribed to a glomus jugulare tumour.

Up to the end of 1956, 172 tumours of the glomus jugulare in the vicinity of the middle ear have been recorded. Only fourteen definitely proven cases were reported before 1945 and the names given to these indicated that the growth was thought to be of vascular origin. The resemblance in histological structure between these neoplasms and the "potato tumour" of the carotid body was noted in 1937 and again in 1945. The origin of the tumour in a jugular glomus body was discovered in 1945 and during the ensuing eleven years, 158 cases have been reported. The majority have occurred in middle-aged women.

In addition to the accepted cases in the literature, others in which the diagnosis was considered are listed in Tables I and II. Some of these probably are glomus jugulare tumours although they cannot be regarded as proven, either because the histological descriptions are inadequate or because illustrations are lacking.

The histological appearances show moderate variation. The characteristic "epithelioid" tumour cells occur in cords or in small, solid masses which often have an alveolar arrangement. The peripheral cells are in contact with the walls of capillaries or blood spaces. Scanty collagenous stroma surrounds groups of tumour cells but not individual cells; reticulin fibres have a similar arrangement. Blood vessels and blood spaces are usually numerous.

Growth of the tumour is slow. No proven case of metastasis has occurred but local invasion is a conspicuous feature and destruction of tissues may be extensive. In seven patients, a tumour of the glomus jugulare in the vicinity of the middle ear has been associated with a coincident tumour in another chemoreceptor organ.

The majority of patients have aural symptoms only. The presenting clinical feature is usually a polyp in the external auditory canal which tends to bleed easily and profusely because of the numerous blood vessels in its substance. Cranial nerve palsies, developing either a considerable time after the onset of aural symptoms or concurrently with them, occur in just under 20 per cent. of cases. In a few patients, neurological features have developed before aural symptoms.

The most common conditions simulating a glomus jugulare tumour are: chronic suppurative otitis media with aural polyp formation; an acute inflammatory process in the middle ear or its vicinity; carcinoma of the middle ear or the external canal; and a primary or secondary tumour of the base of the skull.

Treatment of early cases with limited, accessible growth should be by radical mastoidectomy with removal of all visible neoplastic tissue; radiotherapy may be given as a planned procedure following the operation or may be reserved for a possible recurrence In cases which are more advanced but which show no extensive deep erosion of bone, radical mastoidectomy should be performed with removal of as much tumour tissue as possible, operation being followed by radiotherapy Cases in which the growth has spread extensively by the time the patient is first seen should be treated by radiotherapy alone Radiotherapy probably reduces the vascularity of the lesion, the tumour cells are not considered to be radiosensitive

The slow growth of the tumour and the relatively advanced age of most of the patients should be considered before advising formidable operative procedures One of the patients described in the present paper has been under observation for over nineteen years (the longest follow-up on record for this type of tumour) The second case recorded here has been under surveillance for over seventeen years The third developed the tumour while under observation for deafness, since the growth was very small it was removed by the endaural route

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ACUTE OTITIS MEDIA IN CHILDREN TREATED WITHOUT MYRINGOTOMY— A FOLLOW-UP WITH AUDIOMETRY IN 88 CASES

By J W DIXON (Oxford)*

MYRINGOTOMY has a traditionally established place in the treatment of acute otitis media In more recent years the giving of the sulpha drugs or antibiotics, or both, in adequate dosage, has come to be regarded as indisputably valuable in cutting short the infection and diminishing the incidence of complications However, opinions differ on the exact place of myringotomy in relation with antibiotic therapy Several recent papers from North America (Goodale (1, 2), Rutherford (3, 4), Davison (5, 6)) have expressed decided views on the subject, and these among others and the fact that the habit has grown in this Department of dispensing with a myringotomy, have stimulated this paper

The purpose of the paper is

(1) To bring together the different opinions expressed in English on this subject in recent years

(2) To state our method of treatment without a myringotomy and our reasons for this

(3) To record an investigation of the hearing levels of patients treated in this way and show that our methods are justified by the long-term hearing results

Text-Book Opinions

Stewart and Lumsden in Logan Turner's book (1956) (7), say that all operative measures, intramuscular penicillin, aural drops and shrinking nasal drops should be tried first, but that if pain sufficient to rob the patient of a night's sleep persists, then a myringotomy should be carried out However, if the inflammation appears severe from the beginning, and perforation seems certain, a myringotomy should be carried out with out delay

Bateman in Scott Brown's book (1952) (8), says that pus in the middle ear should be let out by a myringotomy Pus is suggested by severe continued earache and a red bulging drum. If it is thought that there is no fluid in the middle ear, a myringotomy may be withheld, and the case watched. He adds "This is always a difficult decision to make, and in cases of doubt myringotomy should be performed."

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Dingley in Maxwell Ellis's text-book (1954) (9), favours a myringotomy as well as penicillin for all cases with signs of middle-ear tension, such as severe pain, marked deafness and a bulging or pulsating drum. Only cases with merely congestion of the drum would he treat with penicillin and observe without myringotomy.

Wilson (1955) (10) states that in infants a myringotomy should be done in the presence of minimal changes in the drum, but older children he would treat with antibiotics and "myringotomy only if necessary". He favours myringotomy if it is thought that there is any chance of retained fluid in the middle ear.

Watkyn-Thomas (1953) (11) states that when there is exudation alone but no pus in the middle ear, chemotherapy is justifiable (presumably without a myringotomy), but that if pus has formed, the most favourable time for chemotherapy has passed (presumably a myringotomy is advised then). When the drum is perforated and still bulging and discharge pulsates suggesting inadequate drainage, the perforation should be enlarged. He ends by saying "otherwise and when in doubt incise the drum".

McLaggan and Collier (1952) (12), deplore the indiscriminate opening of the drum in mild cases as likely to lead to secondary infection. They would treat such cases conservatively at first with antibiotics, only doing a myringotomy after 48 hours if pain persists, or the presence of pus is suggested by persistent reddening or bulging of the drum. However, they state that a myringotomy should precede administration of sulpha or penicillin if pain, redness, or bulging of the drum are present when the patient is first seen.

Morrison (1955) (13), states that antibiotics if started early, often obviate the need for a myringotomy, but when pus has formed and drainage is not free, shown by persistent pain, bulging drum and fever, a myringotomy should be done.

Ballenger (1957) (14), states "early incision of the ear drum should be practised after bulging has occurred".

Review of the Literature

NORTH AMERICAN VIEWS

(*a*) Those strongly in favour of immediate myringotomy in the treatment of acute otitis media, Goodale (1), Goodale and Montgomery (2), Rutherford (3, 4), Davison (5, 6), Harrison (15), McLaurin (16), Schenk (17), Walsh (18), Rosenwasser (19), Zonderman (20), Boies (21), Meltzer (22), claim that for the best results it is still essential to give adequate and early drainage to the infected fluid in the middle ear by incising the drum as well as giving antibiotics With a myringotomy there is less likelihood of failure to resolve necessitating mastoidectomy and a better chance of normal hearing finally Reliance on antibiotics alone is dangerous Recently they have seen increasing numbers of cases where a mastoidectomy has been necessary to secure resolution of acute otitis media Many of these cases had been treated initially without myringotomy Also they now see more cases of deafness following acute otitis media, most of these having been initially treated without myringotomy They believe that in these cases an exudate has been allowed to remain in the middle ear due to lack of adequate drainage and that this causes a low-grade inflammation leading to adhesion formation, scarring and deafness

(*b*) More conservative papers by Lindsay (23), Heller (24, 25), Struble (26), Brennemann (27), state that a myringotomy should be done for pain or fever persisting after 24 hours adequate chemotherapy Since antibiotics almost always secure prompt relief of these symptoms, a myringotomy is very seldom needed now Also they doubt whether the case in which the drum ruptures spontaneously or is treated without a myringotomy does any worse than the one treated by myringotomy as far as complications and the final hearing result

None of the above papers in either section (*a*) or (*b*) record hearing results in support of their contentions

OPINIONS FROM EUROPE, BRITISH ISLES AND NEW ZEALAND

(*a*) Supporters of early myringotomy are Wilson (28), Lundgren (29), Sohoel (30), Birdsall (31), Patterson (32), Sparrevohn (33), Monkhouse (34), Garrett, Tingey and Wardale (35) All except the last three were speaking of before or in the early days of penicillin Several of these papers record a follow-up of the hearing but in only two of these was an audiometer used They considered that an early myringotomy reduced the incidence of cases coming to mastoidectomy and gave a better final hearing result

(*b*) Conservative treatment was advocated by Hall (36), Mollison (37), Black (38), and Wheatley (39) In Hall's series treated largely without a myringotomy, audiometric follow-up showed minimal hearing losses Black's cases of acute mastoiditis treated with penicillin showed only slight clinical hearing loss finally, and all those with post-aural œdema but without a fluctuant abscess resolved without either myringotomy or mastoidectomy

(*c*) In a discussion at the Royal Society of Medicine in February, 1957 (40), between E N T Surgeons and General Practitioners, the surgeons favoured a myringotomy especially if pain or a bulging drum were not quickly relieved by chemotherapy. It was pointed out that a myringtomy allows a culture to be made and also observation of the increase or decrease of discharge, i.e. the progress of the case. It also reduced pain and the length of the illness dramatically (Taylor, Tresidder, Capps, Bateman). However, Wheatley (General Practitioner) said that antibiotics caused resolution so swiftly that a myringotomy was seldom needed now. He had seen 50-100 new cases each year for 10 years, some perforated already. All are now healed, and have good hearing as determined by whisper tests. Abercrombie and Barker (General Practitioners) said that even in the pre-antibiotic era myringotomy cases came to mastoidectomy as often as non-myringotomy cases.

Friedmann (bacteriologist) said that the huge majority of organisms causing acute otitis media were still penicillin-sensitive.

Method of Treatment Used in This Department

When a patient is first seen with acute otitis media, we treat him with a course of intramuscular crystalline penicillin 500,000 units b.d. for 5 days as well as with vasoconstrictor nasal drops. A myringotomy is not included in our routine. This is the case regardless of whether the drum is bulging or not, and whether there is great pain and pyrexia or not.

We find that it is extremely rare for a case of acute otitis media not to lose its acute symptoms quickly and to fail to clear up when penicillin is given early, regularly, and in adequate doses by the intramuscular route without a myringotomy.

We reserve a myringotomy for pain, pyrexia or a bulging drum if any or all of these persist for 24 hours after commencement of treatment, but we rarely find it necessary.

Similarly if, after the acute stage has settled, there appears any suggestion by drum appearances or persistence of deafness that there is residual fluid in the middle ear, we would let that fluid out by myringotomy. We have only occasionally seen this in cases diagnosed and treated from the first under our direction during the last seven years.

On the other hand we frequently see cases which have not completely resolved after an unspecified quantity of sulpha or oral or intramuscular penicillin given outside the hospital, and which speedily resolve when treated with regular and adequate injections in hospital.

We believe that the important thing in acute otitis media is to get speedy resolution of the infection in the mucosa of the middle-ear cleft, as prolonged infection there may cause scarring and deafness. Penicillin can do this *viâ* the blood stream. At the same time the reduction of the mucosal swelling must reduce middle-ear tension and open the eustachian tube so that any exudate can then drain away. Spontaneous perforation of the tympanic membrane is not by itself the disaster it is sometimes made out to be, and the incision left by myringotomy is just as likely to leave a permanent perforation and deafness if active infection and discharge continue for a long time We believe that it is long continued infection which prevents the perforation from healing, and leaves a high percentage of scarred deaf ears, as has been shown by many authors in the pre-antibiotic era Provided that the infection is quickly controlled, a spontaneous perforation will heal without loss of hearing We believe that properly given antibiotics usually achieve this We must strongly emphasize that however a case of acute otitis media is treated, it must be followed up at regular intervals until complete resolution and a normal tympanic membrane is seen, and, above all, hearing is restored If this is not being achieved, treatment must be changed immediately, and this may be to myringotomy, change of antibiotic (based preferably on culture of discharge), adenoidectomy or simple mastoidectomy

The Method Adopted in this Survey

All the traceable records of patients aged 0-15 years diagnosed and treated in this department for acute otitis media between January 1st, 1950 and December 31st, 1955, were reviewed (i e those aged 0-15 years at the time of their treatment) Those discarded were

(*a*) Those still under 4 years of age and therefore unlikely to give accurate audiograms

(*b*) Those treated with suphonamides—i e a few at the beginning of the period

(*c*) Those who had been diagnosed and treated elsewhere, and only seen at the Radcliffe Infirmary for follow-up

The remainder had been treated with penicillin by injection but without a myringotomy, and these were circularized and asked to attend They included a few cases classified as acute mastoiditis, and treated in the same way, i e with penicillin injections, no myringotomy, and no mastoidectomy Of the 321 asked to attend 88 did so They were seen with their parents, 83 were seen by the author and the remaining 5 by other doctors doing full-time E N T practice In a routine E N T examination particular attention was paid to extracting any history of subsequent ear symptoms, such as earache, discharge and deafness, and in the physical examination, the condition of the tympanic membrane, whether intact, whether scarred, whether mobile or not, was particularly noted An audiometric hearing test was then carried out All these were done on the standard Amplivox machine in a sound-treated room In 57 out of 88 cases these tests were carried out by an audiometrician with 9 years' experience in this kind of work In the remaining cases testing was done by the author in 25 cases, other otolaryngologists in 5 cases, and in one case by a nurse with experience of audiology. In a few children an active or subsiding attack of otitis media was present, and in these the audiogram was postponed for a few weeks.

The Material Reviewed was as Follows

Number of patients = 88 (48 males, 40 females).

Number of ears = 114 (26 bilateral).

Age range (at time of illness) = 4½ months to 14½ years—average age = 6 years.

Average time between otitis media and testing = 4 years 2 months.

Average duration of the disease before treatment begun (from the earliest symptoms whether earache or discharge) = 9·8 days.

Information re any previous ear trouble:

Present in 21 cases	88 cases.
Absent in 13 cases	
No record—54 cases	

Condition of the drum when treatment begun:

(*a*)	Intact	56 ears	114 ears
	Perforated and discharging	58 ears	
(*b*)	Whether bulging or not.		
	Of the intact drums	27 bulging among 56	114 ears
	Of the perforated drums	16 bulging among 58	

Treatment Received

(*a*) All cases were treated with repeated intramuscular penicillin injections. In the majority, i.e. in 58 out of 88 cases, this consisted of 500,000 units of crystalline penicillin twice daily for 5 days. Some of the earlier cases had small doses every 3 hours after a loading dose. Thus 18 cases had 30,000 units 3 hourly and 3 cases had 50,000 units 3 hourly for between 5-10 days after an initial dose of 100,000 units. Five cases had 250,000 units twice daily for 5 days. One case had procaine penicillin 300,000 units once daily for 6 days. One case with post-auricular œdema in addition to 500,000 units crystalline penicillin twice daily for 5 days, had aureomycin 150 mg. every 6 hours for 5 days. Another case had some aureomycin in addition—exactly how much is not recorded. One case had 0·25 gm. of streptomycin intramuscularly twice daily for 4 days as well as 250,000 units crystalline penicillin every 6 hours for 6 days, because of convulsions.

Penicillin injections were given by the Out-Patient Department or by the patient's family doctor, or the District Nurse in

22 cases	22	88 cases
Penicillin injections were given to the patients in hospital in 66 cases	66	

(*b*) All cases were given vasoconstrictor nasal drops
(*c*) A myringotomy was not carried out in any of the cases
(*d*) A mastoidectomy was not necessary to secure resolution in any of these cases
(*e*) Incidence of removal of T's and A's or Ads only, in the interval between otitis media and audiometry, 33 patients out of 88

Results Obtained

1 *Incidence of aural symptoms between treatment of acute otitis media and review*—information about this was obtained in 83 cases out of 88

(*a*) Earache or deafness with cold	15	83 cases
(*b*) Otorrhœa	21	
(*c*) No aural symptoms	47	

2 *Subsequent condition of the tympanic membrane*

(i) Intact	113	114 ears
Dry perforation present (This was a chronic perforation present before the attack of acute otitis media in question)	1	
(ii) Of the 113 intact drums		
Normal appearance and mobility	64	113 ears
Some scarring dullness or lack of mobility	46	
No comment	3	

3 *Audiometric results*

At each frequency these are the average figures expressed to the nearest 0 5 db for each set of cases named The figures are for loss of hearing expressed in decibels compared with the normal Where the audiometer has recorded a figure above the normal level, this has been written down as zero to avoid any exaggeration of the results obtained

4 *Cases coming to mastoidectomy*

In this series of 88 cases (114 ears) treated without a myringotomy no cases needed a mastoidectomy to secure resolution of acute otitis media

It seemed possible that because of the methods of classification and filing, the cases that returned for this review might have given a false idea of the effectiveness of this treatment in clearing their infections without the need for a myringotomy or a simple mastoidectomy So the records of all children aged 0-15 years who had any mastoid operations at all or myringotomy in the years 1950-55 in the Radcliffe Infirmary were reviewed

TABLE I

Audiometric Results for the Various Groups of Ears.

Average time elapsing between treatment for otitis media and audiometry, 4 years 2 months.

	125 cps	250 cps	500 cps	1000 cps	1500 cps	2000 cps	3000 cps	4000 cps	6000 cps	8000 cps
1 Overall Results (114) i e all ears whole series	12·0	7·0	5·0	4·5	3·0	3·5	3·5	4·5	7·0	6·5
2 Condition of the Ear Before Treatment										
A *Drum Intact* (56)	12·0	7·0	6·0	5·5	4·0	4·0	3·5	5·0	7·5	7·5
(*a*) Drum intact and bulge recorded (27)	11·0	6·0	6·0	7·0	5·0	5·0	5·5	5·5	7·0	8·0
A includes (*a*) 3 of those intact later recorded as discharging										
B *Ears Discharging when First Seen* (58) ..	11·0	7·0	5·0	4·0	2·5	2·0	4·0	4·0	8·0	8·0
(*b*) Ears discharging with bulging drum recorded (16)	11·5	8·0	3·0	2·5	1·5	2·0	4·5	4·5	8·0	9·0
B includes (*b*)										
C Ears classified as *acute mastoiditis* (13) (All had post-auricular œdema)	7·0	5·0	2·0	2·0	1·5	1·5	3·0	3·0	7·0	8·5
3 Condition of the Ear at Time of Testing Cases with abnormal drums, i e scars, poor mobility, including perforations (one ear only) 46 ears out of 114 . ..	13·0	8·5	7·5	6·5	4·5	5·0	5·5	6·0	11·0	10·5

In the whole series there were only 4 ears with any marked hearing loss, and these had their audiometric readings for most frequencies between the 20 and 30 db level, the remainder of the readings being better than this level

115 mastoidectomies were carried out during this period The records of 97 were traced The majority were modified radical operations for chronic otitis media There were only 8 cases where a simple mastoidectomy had been necessary to secure resolution of acute otitis media which was known to have been treated with an adequate amount of penicillin by injection, but without myringotomy In one of these discharge continued after mastoidectomy and only cleared after T's and A's were removed Three myringotomies were done during this period for acute otitis media not resolving after adequate penicillin injections given elsewhere These cases included all those whether their initial acute otitis media was diagnosed and treated by us or not and thus they are the few failures from a much greater group of cases treated without myringotomy than the 321 circularized for this audiometric survey We know that a great number of cases of acute otitis media in this region are treated by their own doctors, all without myringotomy, and not referred to us unless complicated However, as this is the main hospital for an area containing at least 300,000 people, almost all cases not resolving or needing operation are referred here

Conclusion

An early myringotomy for acute otitis media has always seemed to the author to be an entirely rational procedure However, the results which were seen in this Department after no myringotomy, and which prompted this investigation, suggest that the operation is very rarely necessary The dangers believed to follow the leaving of infected fluid in the middle ear a moment longer than necessary and so leading to chronicity of infection, increased percentage of mastoidectomies, and poor final hearing, are not borne out Many authors state that the best criterion of success in treatment is the final hearing result, yet very few of those who express decided views about the retention of a myringotomy in the antibiotic era have produced audiometric results to support their points

The above figures would appear to speak for themselves It should be noted that in hospital practice it is the more severe cases that are seen Also the assumption in this paper that all these cases had normal hearing before infection, which is unlikely, serves only to make these average results more impressive An interesting point commented on by others (41) and borne out here is that a scarred tympanic membrane frequently goes with remarkably normal hearing These results were obtained between 1950 and 1955 and it is possible that in those years bacteria commonly causing otitis media were more sensitive than they are now No clinical evidence had been noted yet in this hospital (1957) that this is so and Friedmann's view has been recorded above, but the point needs to be watched

There are many possible fallacies in the collecting of a series such as this. Ideally one should analyse and record the subsequent hearing, in say, any 100 consecutive cases of otitis media treated, but a follow-up based partly upon Out-Patient records does not make this possible. However, it is felt that the absence of relapses and complications and the subsequent high hearing standard of the cases available for study is worthy of record, and strongly supports the contention that the above method without the use of a myringotomy is not only justifiable but desirable. However it should be insisted that cases of otitis media treated by whatever method must be followed until the drum and hearing return to normal, and thus allow of a change in the treatment. Only thus can failures be detected before irreversible damage is done to the sound conducting apparatus and only thus will the so-called masked cases of otitis media and mastoiditis become evident.

Summary

1. Opinions still differ on the place of a myringotomy in the treatment of acute otitis media.
2. The recent literature in English on this subject is reviewed.
3. An audiometric investigation into the subsequent hearing levels of 88 children aged 0-15 years (114 ears) in which acute otitis media was treated with penicillin but without a myringotomy is recorded.
4. Minimal hearing losses were found by audiometry on average over 4 years after treatment. Complications were absent.
5. These results support the contention that the treatment of acute otitis media in children without a myringotomy is justifiable.
6. Adequate follow-up to see that the infection is resolving satisfactorily is emphasized.

Acknowledgments

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CLINICAL RECORDS

A CASE OF UNILATERAL MANDIBULO-FACIAL DYSOSTOSIS ASSOCIATED WITH AGENESIS OF THE HOMOLATERAL LUNG

By T. G. WILSON (Dublin)

MARY F., aged eight years, was admitted to the National Children's Hospital, Dublin, on September 28th, 1954, for advice about a left-sided facial deformity. It was seen that this was due to mandibular dysostosis. On routine examination she proved also to be suffering from absence of the left lung.

Family History

No significant history of disease in her parents, ancestors or numerous collateral relations was elicited. Both her parents and all her brothers are alive and healthy.

Personal History

Mary is the fourth of a family of five. The other four children are boys, ranging in age from 17 to 5 years. None of the boys show any abnormality.

Mary was born on *September 25th, 1946*, at full term after what appears to have been an uneventful pregnancy. She is not an unduly delicate child, and survived measles and whooping cough without difficulty. The "milestones" of post-natal development were attained slowly but within normal time-limits. Owing, it is alleged, to transport difficulties, she did not go to school until the age of seven, and consequently cannot read or write, although she is said to be good at figures and letters and to have an unusually good memory. She can find her own way to school (a distance of a quarter of a mile), knows the way about her town and all the principal shops; plays normally with other children, and is well able to "rough it" with them. In her home she helps with the housework and seems to enjoy doing so. She is, however, shy and timid with strangers, and is handicapped by deafness: total in the left ear, partial in the right.

Examination

1. GENERAL. Mary is a slender child, 3 ft. 8 in. in height and weighs 2 st. 8 lb., and is therefore somewhat subnormal in size for her age. Her I.Q. has been assessed at 75 per cent. after allowance has been made for the deafness from which she suffers.

2. THE MANDIBULAR DYSOSTOSIS. On inspection the face is markedly asymmetrical owing to insufficient development of the left malar bone. In

consequence the palpebral fissure slopes downwards and the whole eye appears to be displaced downwards. No coloboma or other ocular abnormality is present, and the fundi are normal. A fissure extending laterally from the left angle of the mouth causes macrostomia. The chin is deviated to the left side, the deformity being increased on opening the mouth by the unopposed action of the right external pterygoid muscle and the pull of the left depressors, including the digastricus.

On palpation the posterior third of the zygomatic arch and the ascending ramus and coronoid process of the mandible were found to be absent.

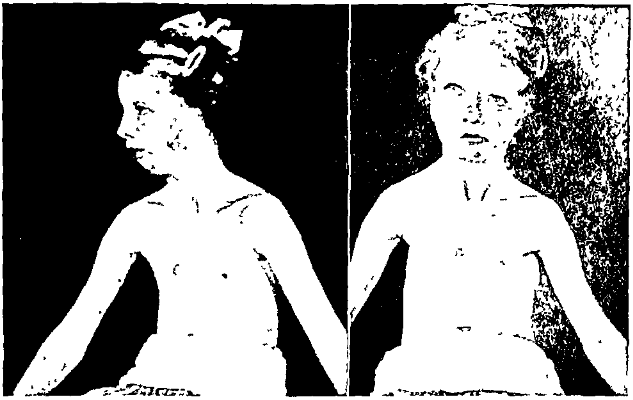

FIG 1

Unilateral mandibulo-facial dysostosis and agenesis of the homolateral lung in a girl of eight years

Intra-oral findings. Mr. Adrian Cowan, M.B., F.D.S., R C S.Eng , reported as follows:

$$\text{Teeth present } \frac{6edch1 \,/\, 10cdeb}{6edc21 \,/\, 12cdeb}$$

$\frac{e/de}{ed/de}$ are very badly decayed and require extraction, $\overline{2|}$ is linguoplaced, and there is marked overcrowding and tilting in the incisor region The arches are small for eight years, and there is a deep overbite and cross bite, $7\overline{|}$ occluding in $\underline{|b}$ area, although the centre between 1/1 is correctly placed in relation to the mandible. The mucosa is normal except for slight chronic marginal gingivitis arising largely from overcrowding in c/c area.

Radiography shows the left ramus and coronoid to be missing from the region immediately posterior to the crypt of the developing $\overline{|7}$. This crypt appears to impinge directly upon the lower border of the ramus, as does the developing distal root of $/\bar{6}$, and a similar arrangement is seen at the right side. Apart from these points, all the permanent teeth are normal in position and stage of development. The third molars are absent, but they do not normally begin to form until some time between the seventh and ninth year.

The Ears

The left ear. The left external ear was found to be grossly deformed and displaced caudo-ventrally. No external auditory meatus was present. A spherical pedunculated mass about ½ in. in diameter, was present at about the normal position of the angle of the jaw From a position about ½ in. below and behind this mass a skin-furrow extended upwards and backwards to the point where the root of the zygoma would normally be situated. This furrow was covered with thin skin immediately underneath which could be felt a thin plaque of cartilage resembling the anti-helix.

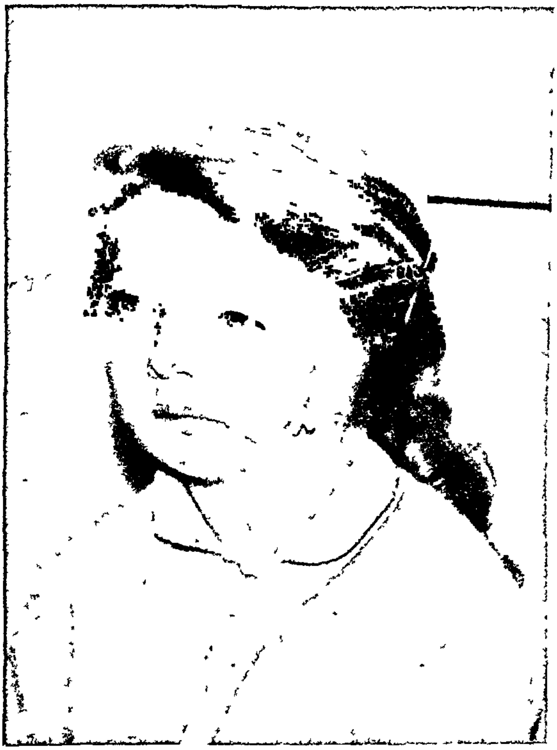

FIG 2

To show the left-sided mandibulo-facial dysostosis

The right ear. The right external ear was normal in appearance and position. The external auditory meatus was anatomically normal but contained a quantity of cerumen and dried purulent debris which when removed disclosed a large central perforation. The ear dried up completely with conservative treatment.

Hearing tests were difficult to carry out and assess, for Mary, while willing and anxious to co operate, did not fully understand what was required of her She responded to conversational voice at about 2 yards and to the whispered voice at a distance of about 4 feet These results and the audiogram which was obtained may show more deafness than is actually present It was, however, clear that she had little hearing on the left side and was considerably deafened in the right ear for both high and low tones As her mother said, probably influenced by a misconception, "she has only half her hearing" She has now (*February, 1957*) been fitted with an electrical hearing aid from which she derives considerable benefit

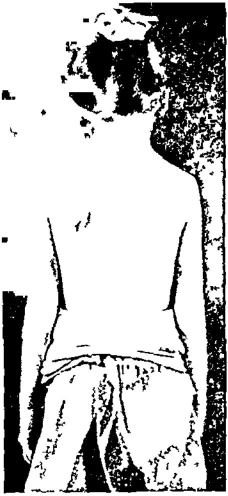

FIG 3
To show the winging of the scapulae

Balancing tests. Caloric tests were approximately normal for the right ear. Rotation tests confirmed this and gave a normal response on the left side

3 THE PULMONARY AGENESIS Inspection of the thorax showed marked retraction of the ribs on the left side forming a sulcus extending from the clavicle to the left hypochondrium Winging of both scapulae was present Further examination of the thorax showed that the left lung was absent the left side of the thorax being occupied by the heart and part of the right lung Bronchoscopy revealed that the trachea, which narrowed as it approached

the bifurcation, was deflected towards the left side of the thorax. At the bifurcation the right bronchus appeared to branch off to the right, while the rapidly narrowing left bronchus went straight on and ended blindly in about 3 cm. Radiography confirmed these findings and showed that the left bronchus ended in a small mass of tissue which represented the unexpanded left lung. The absence of the left lung was confirmed by radiography and bronchoscopy.

Further examination disclosed that the sixth cervical vertebra was a hemi-vertebra, the defective portion being on the left side, and small cervical ribs were present on both sides. Apart from this no more abnormalities, visceral or skeletal, were found.

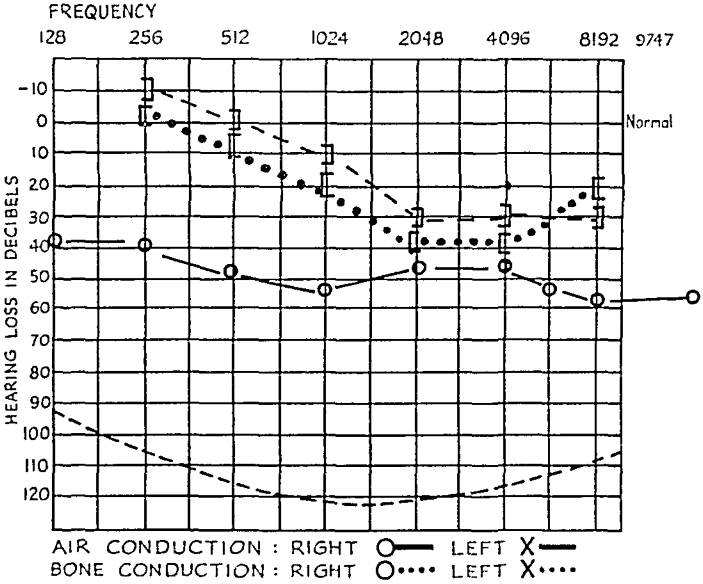

FIG. 4.
M. F's audiogram.

Radiography by Dr. Sholto J. Douglas gave the following results:

Skull. There is failure of development of the ascending ramus of the left mandible. The posterior half of the left zygomatic arch is missing.

The right mastoid process appears reasonably developed. The right mastoid cells are cloudy, probably due to infection. The inner-ear development is normal on both sides, as also are the semi-circular canals.

The left mastoid is very poorly developed as compared with the right. Both optic foramina are normal.

Spine. The sixth cervical vertebra is a hemi-vertebra, the deficiency being on the left side. Small cervical ribs are present on both sides. The remainder of the spine is normal.

Neck. Calcified glands are present in the right side of the neck.

Chest. Opacity of the left side of the chest appears to be due to agenesis of the left lung There are gas shadows ascending high into the left side of the thorax suggesting that the stomach is situated in the left thorax (but see below).

Bronchogram There is a rudimentary left main bronchus with a small rudimentary lung bud shown filled on the left side The right lung is much displaced across the midline

Barium Meal A barium meal does not show any evidence of diaphragmatic hernia and the left diaphragm appears normal Many small opacities are present in the abdomen which appear to be beads and some of which are in the appendix

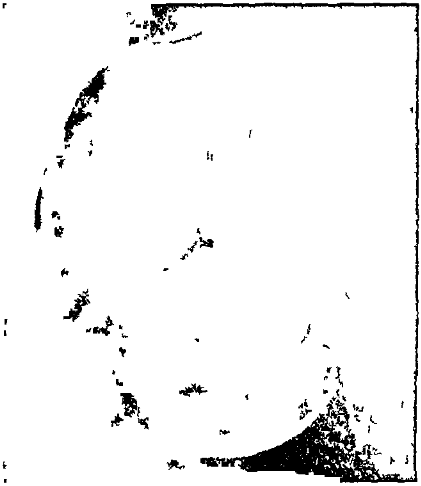

FIG 5
Absence of ascending ramus of left mandible and poor development of zygomatic bone

Intravenous Pyelogram Both kidneys are normal and the bladder appears normal

Arteriography was carried out by Mr John Lanigan F R C S I , at St Laurence s Hospital The left internal and external carotid systems were apparently normal

Discussion

Mandibulo facial dysostosis has been classified by Franceschetti and Klein (1949) into complete, incomplete, abortive, unilateral and atypical forms but the types overlap considerably and it is not possible to divide the cases into exact groups The present case is obviously unilateral, and is incomplete in that

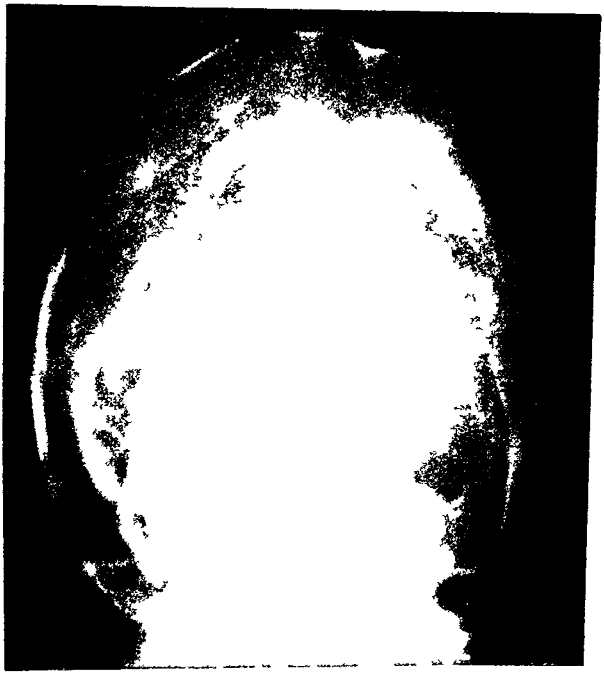

FIG 6
Failure of development of left mastoid

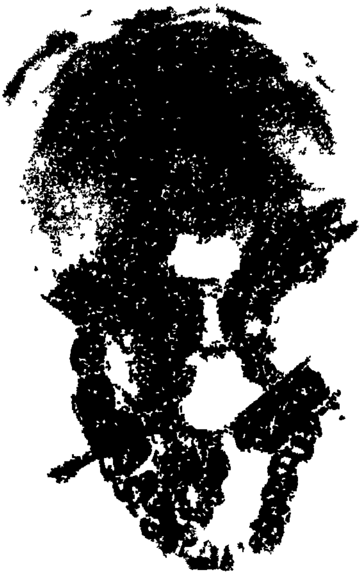

FIG 7
Abnormal left mandible and absence of mastoid process on the left side

o colobomata are present the only ocular abnormality present being the iti mongoloid obliquity of the left palpebral fissure

According to Franceschetti and Klein the condition is due to an inhibitory rocess occurring towards the seventh week of embryonic life and affecting the cial bones deriving from the first visceral arch It is of genetic origin and ansmitted as an irregular dominant being an unstable gene of occasional leiotropic (polyphenic) effect especially on the skeletal system As regards ne etiology we have to assume a disturbance of the action of the organization

[illegible] 8

Absence of upper portion of [illegible] dible normal orbit and external ear

entre which is probably due to a lethal or sublethal effect of the gene concerned (*loc cit*)

McKenzie and Craig (1955) arguing from a case which they have dissected have put forward the suggestion that the condition results from a defective stapedial artery which they say causes maldevelopment not only in its own field of supply but also in the region of the first visceral arch whose vessels the stapedial artery normally supports during the critical phase between the disappearance of the first aortic arch and the full development of the external carotid artery just after the formation of the primitive face

They make the interesting suggestion that instead of the supraorbital infraorbital and mandibular arteries arising from a single point on the first aortic arch they arise separately and that the infraorbital which originally runs deep to the mandibular nerve, is the vessel which receives the stapedial artery and depends most upon it for survival. Without the help of the stapedial artery the middle meningeal artery could survive by retaining its connection with the dorsal aorta, and similarly the mandibular artery could continue to function through its connection with the ventral aorta, which becomes the external carotid. However, this attractive theory must in our opinion be condemned for various reasons, including the fact that it does not explain the frequent occurrence of skeletal defects in the cases described in the literature.

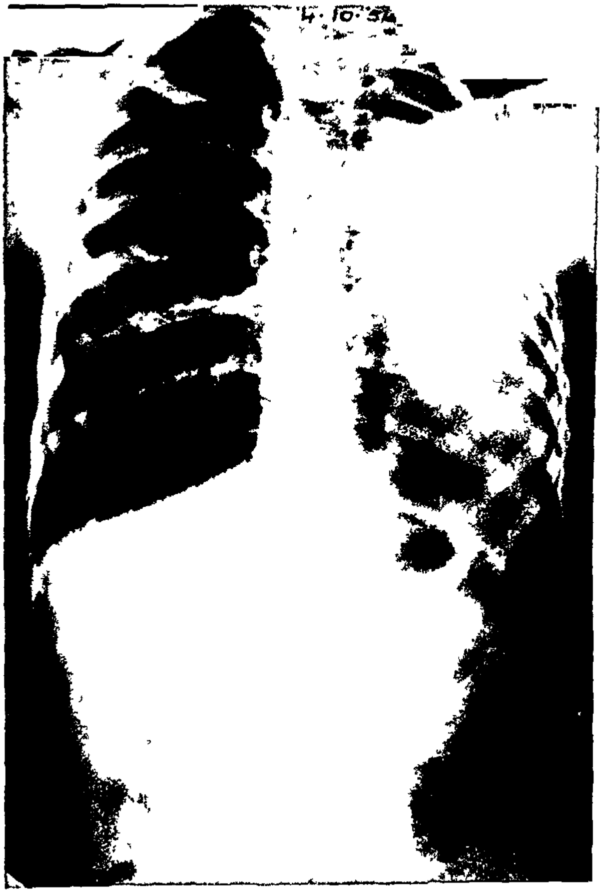

FIG 9
Agenesis of left lung and displacement of right lung

Agenesis of the lung is divided by Schneider (1912) into three groups of cases. They are (1) Total absence or true agenesis, in which there is no trace of lung, bronchus, or vascular supply to the affected side, (2) cases in which a primordial lung bud is present, as represented by a tiny, out-pocketing of the trachea, but no lung tissue, and (3) those in which a narrow bronchus ends in a fleshy structure which lies in the mediastinum representing the lung The present case falls into the third category It is of interest that of 87 cases collected by Per Wexels (1951) only twelve cases reached the age of eight years or over five of these being diagnosed during life—which puts the present case and the second reported below into a very select group It is however probable that many more cases reach adult life It is also pertinent that one of the 87

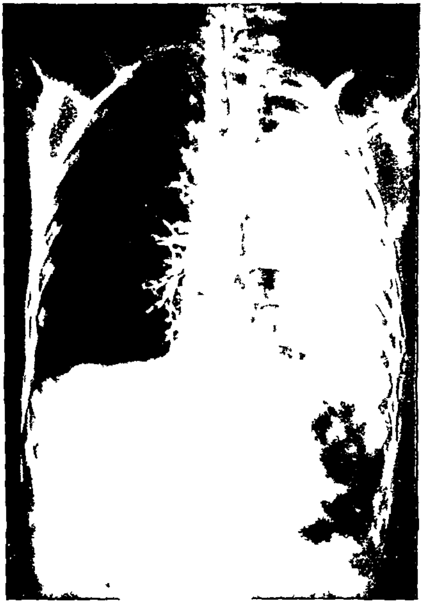

FIG 10

Bronchogram showing a rudimentary left main bronchus with a small rudimentary lung bud shown filled on the left side The right lung is much displaced across the midline

a boy who died at the age of six months is described as having suffered from a deformed external ear while another had hypoplasia of the face which might have been dysostosis cleidocranialis Crouzon s disease (hereditary craniofacial dysostosis) or dysostosis mandibulo facialis as in the present case In all nearly 50 per cent had other congenital defects usually of a major nature This leads one to believe that the condition is a true malformation and probably the result of an inherent defect in the germ plasm (Ferguson and

Neuhauser, 1944). In contra-distinction to mandibulo-facial dysostosis, agenesis of the lung appears to occur most often on the left side, as in this case

According to Spemann (1936) the normal development of the whole body takes place under the inductive influence of the so-called organizers These are, generally speaking, areas of the embryo which exercise a specific regulatory influence on morphogenesis by producing certain chemical substances (evocators) which bring about the differentiation of the various organs and tissues. The primary organizer material is derived from the dorsal lip of the gastrula. It gives rise to a head organizer, which induces the differentiation of head, brain and sense organs, and to a trunk organizer which induces the differentiation of the trunk. In the course of this embryogenesis, organizers of lower grade (secondary, tertiary, etc) are formed Development obviously depends on the presence of normal organizers if the latter are damaged, faulty development of the affected areas will result

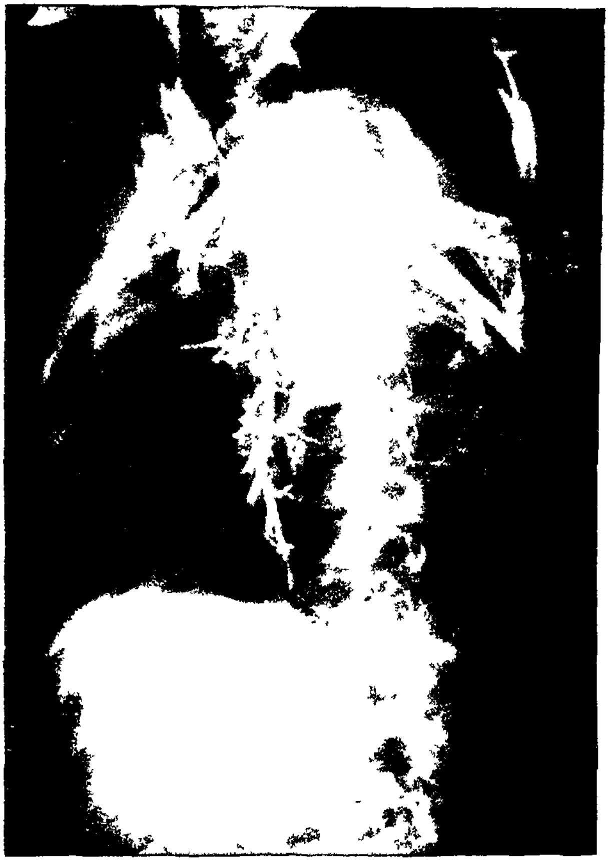

FIG 11

Oblique film of bronchogram showing normal right bronchus

Damage to the organizers may occur in various ways Gene mutations occur fairly frequently in mammals, including man The abnormal gene exercises a specific effect on a certain area in the case of mandibulo-facial dysostosis it is principally the organizer of the head which suffers If another malformation is constantly associated with another, as for instance mandibulo-facial dysostosis is with defects of the radius, gene-linkage is probably the cause, particularly if the deformities are hereditary Sometimes, however, a single pleiotropic gene may cause a widely varying series of congenital defects, and in these cases it is likely that the fundamental anomaly is metabolic and may manifest itself in different ways (Neeham 1942)

However, as Altmann (1949) says, not all congenital syndromes are genetically determined Environmental interference with the early developing embryo may have similar results, and repeated cases may be due to repeated exposure of the embryo to the same harmful environment Gillman (1948) has shown experimentally that injection of trypan blue into rats can cause atresia of the ear amongst many other deformities Other chemicals have an injurious effect on the later stages of development Infection may have the same effect, as is well known in the case of maternal rubella Vitamin A deficiency in experimental animals can produce major developmental defects as also can riboflavin deficiency Irradiation is another cause of hereditary changes in the offspring, but this is regarded as "the result of reciprocal gene translocation rather than gene mutation ' (Snell, 1941 Hertwig 1944) Finally, mechanical factors *in utero*, malposition, hydraulic pressure, and constriction by amniotic bands are said to cause congenital defects, but while one can understand how such conditions as talipes equinovarus or congenital dislocation of the hip could be caused in this way it is difficult to understand how it could give rise to mandibular dysostosis or atresia of the lung

It is probable therefore that this case is caused by a disturbance of the head organizer by an unstable gene of pleiotropic effect The unexplained features are the strictly unilateral nature of the lesion—unilateral mandibulo-facial dysostosis is quite a rare condition—and the complete absence of a significant family history

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AGENESIS OF THE LUNG IN A BOY OF TEN YEARS

By T. G. WILSON (Dublin)

EARLY in *January, 1957,* a boy aged ten years and four months was brought to the Out-patient Department of Dr. Steevens' Hospital, Dublin, by his mother, who complained that he suffered from frequent colds with nasal obstruction and discharge. She further stated that he had only one lung.

Family History

No significant family history was elicited. The mother is Rh. negative.

Timothy is the second of five children, his elder sister, who is said to be

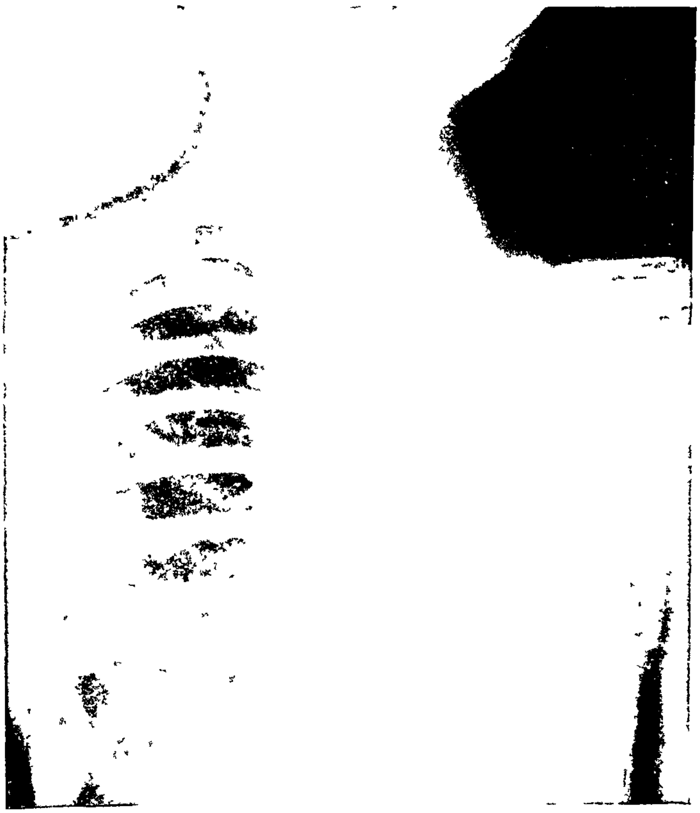

FIG. 1

The side of the chest is opaque and the rib-spaces are diminished. The trachea is displaced to the left. The level of the diaphragm on the left side is not shown. The heart is displaced markedly to the left. The appearance is that of agenesis of the left lung.

quite normal and healthy, having been born in December, 1946 Two boys and a girl were born subsequently, all of whom died of kernicterus in spite of active treatment including whole blood transfusions

Examination

Both nasal cavities were congested and obstructed and contained mucopus The tonsils were enlarged and the lymph nodes in the submaxillary triangle were swollen on both sides General physical examination appeared to confirm the mother's statement that the left lung was absent

X-ray studies of the chest were carried out, Dr Sholto Douglas reporting as follows

> *Chest* The side of the chest is opaque and the rib spaces are diminished The trachea is displaced to the left The level of the diaphragm on the left side is not shown The heart is displaced markedly to the left The appearance is that of agenesis of the left lung Tomographic sections confirm this impression, but also suggests that there is a herniation of part of the right lung through the upper mediastinum
>
> There is a congenital deformity of the left lower cervical spine with the formation of a hemi vertebra at the level of the VIIth cervical There is a rudimentary first rib on the left side

It was arranged that the child should be admitted to hospital a fortnight later for bronchoscopy and bronchography, but he did not arrive on the date arranged, and no reply was received to a letter addressed to his parents

Comment

This obviously incomplete account is reported merely in order to put the case on record The patient falls into Schneider's type III and his case is in fact very similar to the previous one except that the only associated congenital abnormalities are the hemi-vertebra and a cervical rib, both of which are also present in the first case

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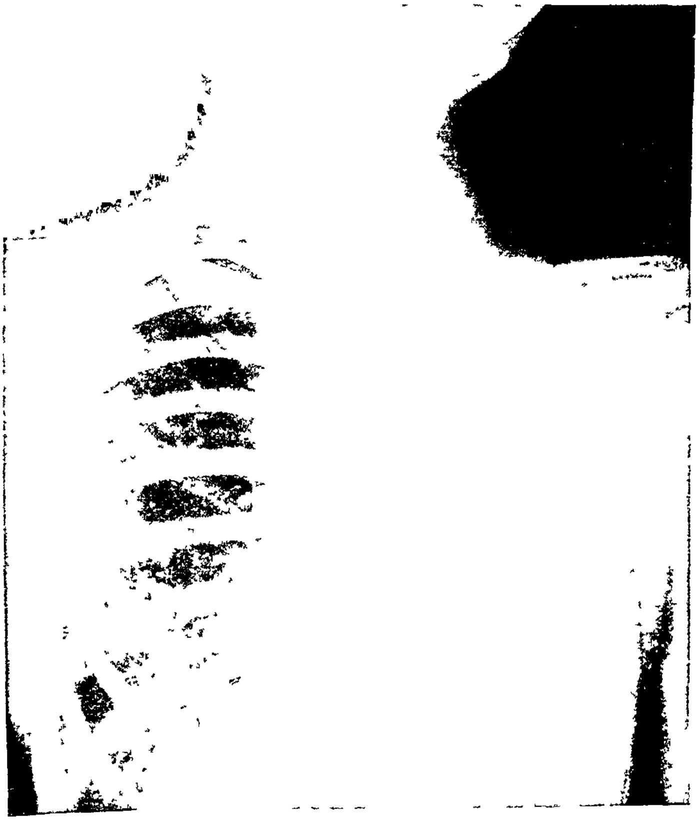

Fig 1

The side of the chest is opaque and the rib-spaces are diminished The trachea is displaced to the left The level of the diaphragm on the left side is not shown The heart is displaced markedly to the left The appearance is that of agenesis of the left lung possess a strong anti emetic action together with some of the characteristic properties of its related compounds (Ducrot and Koetschet 1956) Its formula is given as 1 (3 (3 Chloro 10 phenothiazinyl)propyl) 4 methyl piperazine

The four severe cases of Meniere s disease in this trial had been previously tried with dramamine and avomine (amongst other drugs) and while they seemed to lessen the severity of the vertigo they did not possess the ability to cut short or abolish major and minor episodes of vertigo which was the case with Stemetil All three drugs act on the so called vomiting centre to a greater or less degree (Broussolle and Dubor, 1956), but Stemetil being allied to chlorpromazine has a slight hypotensive action and to a lesser extent hypnotic and temperature lowering effects indicating some central vasomotor or hypothalamic level of activity Goodman (1957) argues that the treatment for Meniere's disease should be directed to the smooth muscles of the arterioles rather than to the end product of the disease endolymphatic hydrops It would appear that Stemetil has a greater action on the vertigo produced by a labyrinthine vascular upset than it has on the vertigo of otosclerosis arterio sclerosis or virus diseases of the vestibular system in which conditions the pathology is relatively static compared with the dramatic variability of endolymphatic pressure which occurs in Meniere s disease A series of patients with migraine assessed elsewhere with satisfactory results (excluding pre menstrual cases) seems to lend support to the idea of an action of Stemetil on the cerebral vascular system In both of these conditions the subjective result of the administration of the drug was probably enhanced by virtue of its relaxant and sedative effects (Broussolle and Dubor 1956)

This is a difficult disease to assess and the sufferers tend to be over-enthusiastic about any new remedy, especially the ones with bilateral disease or who are over the age of 55 when cervical sympathectomy is not safe (Lewis and Harrison 1956) No real conclusions can be drawn from the study of four severe cases but so far Stemetil has proved to be of far greater value than avomine, the antihistamines nicotinic acid or phenobarbitone in severe Menière's disease It is to be hoped that a wider trial will be made with Stemetil over a long period of time in order to give it an established value in this distressing disease

Brief summary of the four severe cases

CASE 1 Female aged 62 Four years history of attacks of vertigo, vomiting and deafness Sometimes weeks, sometimes days between the attacks, but with frequent transitory vertigo Tinnitus constant The ears show no sign of previous middle ear disease There is a perceptive deafness in both ears with a threshold at 60 db at 8 000 c p s This patient is overweight

CASE 2 Male aged 66 This man has severe osteoarthritis and he has had twelve major attacks in ten years lasting two hours or more Between these severe attacks he has frequent small bouts of vertigo when lying down or on moving about There is scarring on the right tympanic membrane and the mobility is reduced There is no fluid in the middle ear He has a right perceptive deafness with a threshold at 80 db at 8 000 c p s

CASE 3 Female aged 55 In the past ten years this woman has had severe attacks of vertigo which have increased in number to four in the past year She has a history of discharging ears and both tympanic membranes have less than normal mobility. She has a bilateral perceptive deafness most marked on the left side where the threshold is at 100 db. at 8,000 c.p.s.

CASE 4. Female, aged 59. In seven years this patient has has fifteen major episodes. Between times she says she has had numerous attacks of a minor character. Again she has a history of discharging ears in the past and on the right side there is a reduced mobility of the tympanic membrane. There is a bilateral perceptive deafness more marked on the right side where the curve drops to 60 db. at 8,000 c.p.s.

In these four cases the audiographs have varied from time to time, sometimes by as much as 30 db. Also the caloric tests have varied a great deal and because of this they are not reproduced.

Acknowledgment

I am grateful to Messrs. May & Baker, Ltd., for the supplies of Stemetil used in this trial.

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LABYRINTHOGENIC MENINGITIS DUE TO BACILLUS PROTEUS

By B H COLMAN (Edinburgh)

MENINGITIS of labyrinthine origin appears to be increasingly uncommon, furthermore as this case presented a number of interesting points it would seem worth recording

Cholesteatoma is encountered in a considerable proportion of cases of chronic suppurative otitis media of attico antral type and use of the microscope routinely in middle ear dissection not infrequently reveals shimmering crystals of cholesterol even when there is no suggestion whatever of cholesteatoma clinically

Before cholesteatoma has had the opportunity to demonstrate its dangerous potentialities and full destructiveness however, increasing deafness and fœtid discharge usually cause the patient to seek expert advice The following case is exceptional in that disease was allowed to progress for years in a patient who was much too busily occupied to be inconvenienced by minimal symptoms virtually unchanged since boyhood until a sudden (and almost disastrous) intracranial complication brought him to hospital

The patient was an intelligent and co operative miner of 32 employed at the coal face There was a history of occasional slight discharge from the right ear with associated deafness For three weeks prior to admission there had again been a trace of discharge for which he had received a daily injection of penicillin from his own doctor There was discomfort over the mastoid for a week and for thirty six hours there had been severe occipital headache with vomiting He nevertheless continued at work and was actually on the way down the pit to commence another shift when he collapsed

On examination, the signs were those of a severe meningitis The left ear was full of cholesteatoma and appeared to be completely disorganized, the meatal wall was sagging considerably, he was stone deaf on this side and the fistula sign was negative There was no nystagmus present the labyrinthine destruction appeared therefore not to be recent Slight weakness of the right side of the face was observed Lumbar puncture was carried out with the following results 220 mm pressure, very turbid fluid with free rise and fall on jugular compression, cell count 13 000 c mm (mainly polymorphs), sugar 12 mg per cent , protein 146 mg per cent chloride 668 mg per cent Treatment was instituted along conventional lines with the administration of systemic penicillin, streptomycin and sulphadiazine as well as intrathecal penicillin and streptomycin whilst awaiting the bacteriological reports These revealed the presence of proteus vulgaris (fortunately sensitive to streptomycin) in the ear and in the cerebrospinal fluid An X ray taken at this time is illustrated and shows very extensive bone destruction (Fig. 1); the opposite mastoid was fully pneumatized.

Improvement on the above chemotherapy was extremely rapid, within ten days he appeared clinically well and with a cerebrospinal fluid not far from normal. On the twenty-first day after admission it was decided to allow him home on sulphonamide cover (his third course) in order to permit 7-10 days recuperation amongst his family prior to re-admission for appropriate surgical treatment, the cerebrospinal fluid was normal and the only treatment in the last week had been systemic penicillin in moderate dosage. On the day of departure a cold caloric test was carried out to discover whether any labyrinthine function remained, no nystagmus was elicited.

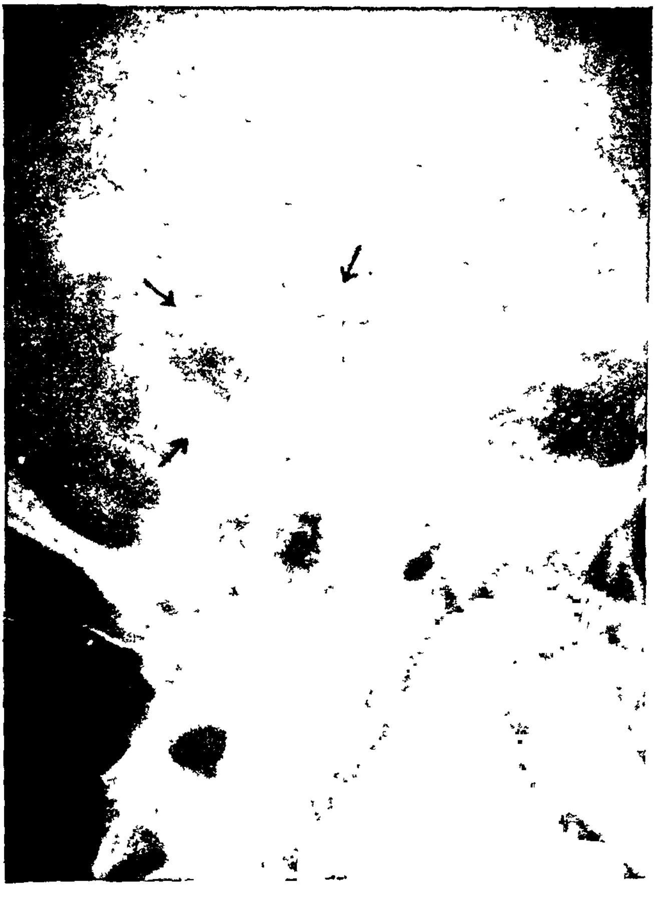

FIG. 1.

Extensive bone destruction due to cholesteatoma.

Within twenty-four hours the patient was re-admitted with a recurrence of his meningitis and a cerebrospinal fluid cell count of 8,000 c.mm. Fortunately, response to treatment was again rapid though it was felt that the meningitis was no more than controlled by chemotherapy No organisms were isolated this time, but the cell count never fell below 250 Operation was therefore decided upon eight days later

Exploration confirmed what the radiographs suggested almost the entire mastoid had been destroyed, the cholesteatomatous mass being retained superficially only by periosteum of the cortex and of the postero-superior wall of meatus Middle and posterior cranial fossa dura was uncovered and the sigmoid sinus widely exposed Gentle removal of the mass anteriorly showed that the facial nerve was exposed from the region of the geniculate ganglion to about the middle of the descending portion and in part its course was completely surrounded by the cholesteatoma The promontory was intact but there was no trace of the lateral semicircular canal, only a hole leading into the vestibule

Adequate labyrinthine drainage was obtained by removal of the promontory, after which a probe was easily passed between the superior and inferior parts of the labyrinth in the usual way medial to the facial nerve (now lying unsupported over a considerable length)

Post operative progress was uneventful The very large cavity was Thiersch grafted after a week and the patient went home a fortnight later completely well

Comment

This is a fairly typical case of meningitis of otitic origin but nevertheless presents several interesting features and illustrates some old but valuable lessons

1 The patient had a chronic suppurative otitis media for years, yet in spite of the extent of the cholesteatoma he had minimal symptoms There was nothing in the past history to suggest when labyrinthine destruction occurred, neither was he aware of the fact that the ear was stone deaf

2 The case illustrates the hazard to which a patient harbouring a dead infected labyrinth with full compensation is exposed He is in constant danger of meningitis and as the labyrinth is already functionless there is no warning labyrinthine storm to suggest the otitic origin

3 The recurrence of meningitis immediately after performing a cold caloric test emphasizes the danger of this simple procedure in patients recovered from labyrinthogenic meningitis

The test was done in this case not out of curiosity, but because a short time previously labyrinthine drainage had been carried out on the stone deaf ear of a somewhat similar case and rather surprisingly was followed by nausea and paralytic nystagmus lasting two to three days In spite of this, it is felt that in these circumstances examination with a Barány noise box is sufficiently reliable in diagnosing absence of labyrinthine function for practical purposes

4 The organism in the cerebrospinal fluid was unusual Bacillus proteus is an extremely uncommon intruder even though frequently present in chronic middle ear suppuration as a secondary invader Fortunately it was sensitive to streptomycin The last case of proteus meningitis seen in this department was in 1942 and in the absence of antibiotics was attended by a fatal result (Sugar, 1944)

5. The operative findings well demonstrate the remarkable extent to which cholesteatoma erodes hard bone yet is arrested by soft tissue planes. Even the facial nerve, though entirely surrounded in part of its course, was completely intact. The temporary facial weakness pre-operatively was undoubtedly the result of a sudden increase in pressure associated with the exacerbation of infection.

I wish to record my thanks to Dr. Simson Hall for permission to report this case and to Mr. Paterson of the Wilkie Surgical Research Laboratory for having prepared the photograph.

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GENERAL NOTES

THE Editors are anxious to improve the clinical aspect of the Journal and would welcome more reports of interesting cases These clinical records need not be at all elaborate and it would be unnecessary to go minutely into the ætiology or the literature of the subject Senior Registrars have unique opportunities for seeing a large variety of cases, many with unusual features It is in many ways regrettable that these should go unrecorded It is hoped that all those in charge of clinics will help us in this matter and see that such cases are not overlooked

OTOLARYNGOLOGY LECTURES 1957-58

THE following lectures, arranged jointly by the Royal College of Surgeons of England and the Institute of Laryngology and Otology, will be delivered in the Lecture Hall of the College, at 5 30 p m

1958
May 1st Professor W D M Paton The pharmacology of mucous membrane
July 3rd Mr Norman C Tanner Haemorrhage from the Œsophagus

INTERNATIONAL CONGRESS ON THE MODERN EDUCATIONAL TREATMENT OF DEAFNESS

AN International Congress on the Modern Educational Treatment of Deafness will be held at the University of Manchester from July 15th-23rd 1958

The Congress is being organized with the approval and support of the Council of the University, with financial help from the Ministry of Education the National College of Teachers of the Deaf, and the Rathbone Trustees and with the co operation of the British Council and the Alexander Graham Bell Association for the Deaf, Inc , Washington, D C , U S A

The Congress is being planned to bring together educators otolaryngologists, pædiatricians, medical officers audiologists and educational psychologists, to consider and discuss the significance, to the future development of their work, of the results of modern research on problems of deafness in childhood

Arrangements are being made to offer accommodation to members of the Congress in University Halls of Residence The cost of this type of accommodation is expected to be about £1 2s per day, including bed breakfast and dinner The University Refectory will be available for meals during each day

Forms of application for membership of the Congress will be supplied upon request, to Professor A W G Ewing, Department of Education of the Deaf, the University, Manchester 13, England

General Notes

EXAMINING BOARD IN ENGLAND

BY THE

ROYAL COLLEGE OF PHYSICIANS OF LONDON

AND THE

ROYAL COLLEGE OF SURGEONS OF ENGLAND

DIPLOMA IN LARYNGOLOGY AND OTOLOGY

THE Secretary reports to the two Royal Colleges that at the First Part of the Examination held in December, 1957, 49 Candidates presented themselves, of whom 26 passed; and that at the Second Part of the Examination held in December, 1957, 33 Candidates presented themselves, of whom 21 passed.

The following 21 Candidates, having complied with the regulations of the Board, are eligible to receive the Diploma, viz:-

Name	Qualification	Address
*Abbey, Paul	M B , S B London (London)	40, Ashburn Place, S W 7
Ahmad, Rasheed	M B , B S Karachi	201, Bhurgri Road, Hyderabad, Sind
Alvi, Zaheerul Haque	M B , B S. Karachi	Sussex Throat and Ear Hospital, Brighton 1
Bihari, Julian	M.B , B S London L R C P , F R C S (U C H)	19, Fieldend Road, Streatham, S W 16
Cartmill, Thomas	M B , Ch B Glasgow	456, Victoria Road, Glasgow, S 2
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General Notes

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9.30 a.m. Scientific Meeting: Royal College of Surgeons of Edinburgh, Nicholson Street. *Discussion:* "Acoustic Neuroma". Professor Norman Dott. Dr. C. S. Hallpike. Dr. D. Dix.

1 p.m. Luncheon for Members and Guests. Carlton Hotel, North Bridge. Price 15s. inclusive of wines and gratuities.

Afternoon. Garden Party by invitation of the Presidents.

7.15 for 7.45 p.m. Dinner in The Royal College of Surgeons of Edinburgh. Tickets Two guineas each inclusive of wines.

Saturday, June 28th.

9.30 a.m. Scientific Meeting: Royal College of Surgeons of Edinburgh. *Discussion:* "The dysphonias and the scope of speech therapy and their treatment". (Illustrated by recordings). Mr. Maxwell P. Ellis. Mr. L. Willmore.

12.30 p.m. Meeting ends.

For details of accommodation write to the Secretaries of the Sections.

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The Journal of
Laryngology and Otology

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April 1958

STAPES SURGERY FOR OTOSCLEROTIC DEAFNESS

By SAMUEL ROSEN, M.D.* (New York, N Y)

MODERN stapes mobilization for the restoration of hearing in otosclerotic deafness is now nearly six years old. In these years, new developments in stapes surgery have been presented; new horizons have opened up in the surgical treatment of otosclerosis and new problems have arisen to challenge otologists and physiologists all over the world. It is with some of these developments, achievements and problems that I should like to deal.

The procedure first described in 1952 (1) consisted in mobilizing the rigid footplate of the stapes indirectly, by pressure against the stapedial neck with a specially devised mobilizer (Fig. 1). By this method, hearing has been restored to various levels up to normal.

When the footplate is mobilized by this method, the ossicular chain remains intact and its transformer action, described by Helmholtz in 1863 (2), is restored.

As we know, Helmholtz's concept of the sound transformer action of the ossicular chain has been accepted for nearly 100 years as the necessary condition for normal hearing. But as stapes surgery has evolved, new phenomena have been observed.

It frequently happened that during attempts to mobilize the stapedial footplate by pressure at the neck, the crura fractured because of excessive fragility or extreme footplate fixation. Even when the crura resisted fracture, it was sometimes impossible to mobilize the footplate by pressure at the neck. It therefore became necessary to go to the footplate itself, in order to mobilize it directly at the site of the pathological fixation. This method of direct footplate mobilization was first reported in 1955 in *Acta oto-Laryngologica* (3). It consists in wedging a sharply-pointed explorer gently between the rim of the oval window and the periphery of the footplate at its anterior and inferior edges, or at the posterior or superior margins (Fig. 2). When the rigid footplate is pried loose from the otosclerotic bone, its mobility is restored and the hearing improves. Hearing has been restored by direct footplate mobilization to various levels, including normal and near-normal, whether or not the crura were previously fractured. The following case is illustrative of near-normal hearing achieved by this method, which has been maintained for 33 months to date: A 35-year-old woman had bilateral progressive deafness for over

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* Read before the Royal Society of Medicine, London, December 6th, 1957

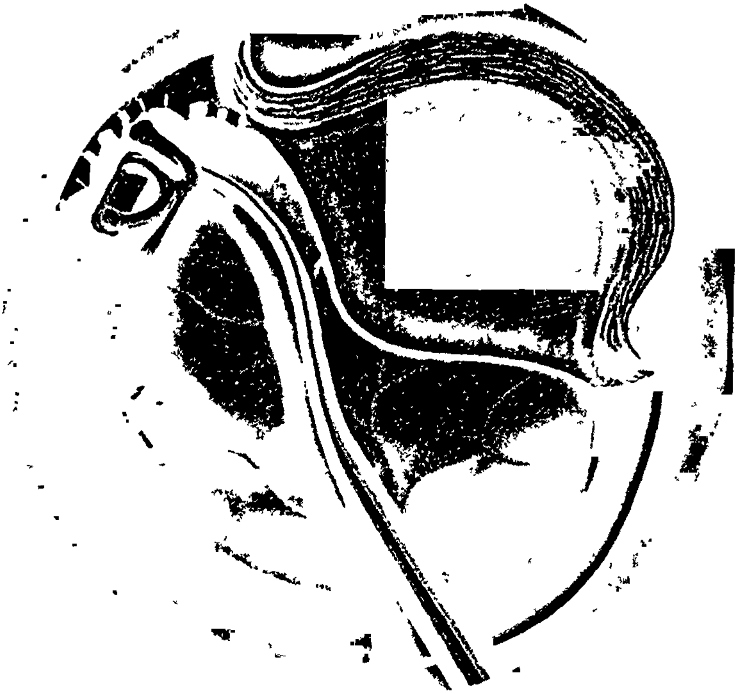


FIG 1.

Mobilization of the footplate of the stapes by the indirect method, with mobilizer exerting pressure against the stapedial neck The drum has been lifted out of its bony sulcus and reflected upward Two-three mm of the posterior bony canal wall have been scraped away to permit full exposure of the stapes and its footplate

5 years Her pre-operative loss was 55 db In March, 1954, a mobilization operation was performed on the left rigid stapes by the indirect neck method The hearing did not improve on the table and 13 months later the hearing was at the 40 45 db level In April, 1955 the same ear was re-operated At the second operation, no technical difficulties or structural changes were found in the external canal or the middle ear Very slight pressure on the neck of the stapes revealed free movement of the stapedial tendon, which was due to fracture of the crura at the initial operation Direct footplate mobilization was then performed The hearing improved on the operating table At her last hearing test two weeks ago, her hearing was still at the 10 db level shown here (Audiogram, R C , Fig 3)

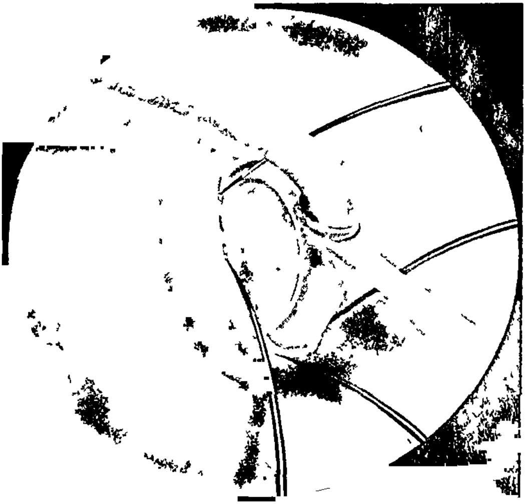


FIG 2

Mobilization of the footplate directly at the footplate The explorer penetrates the oto sclerotic bone at the periphery of the footplate at any of the sites shown here

As more experience was gained with stapes mobilization at the neck and at the footplate, new problems naturally arose We encountered footplates with extensive ankylosis and crura reduced to extreme fragility by otosclerotic involvement Since in many of these cases the footplate could not be mobilized it seemed logical that sound would have to enter the cochlea at the site intended by nature, by way of an opening through the rigid footplate into the vestibule.

In August, 1954, this technique—fenestration of the oval window—was first employed. It was reported at the A.M.A. Convention in Chicago, in June, 1956, and subsequently in the *Archives of Otolaryngology* (4). The fenestra ovalis through the rigid footplate is made with either a crochet-hooked instrument called the oval window fenestrator, or the sharp-pointed explorer (Fig. 4). The instrument penetrates the rigid footplate in its posterior half for not more than a millimetre, either at the periphery or through the body of the footplate as close to the junction of the posterior crus as possible (Figs. 5, 5a, 5b). The point of the instrument is directed posteriorly, away from the utricle and saccule. After

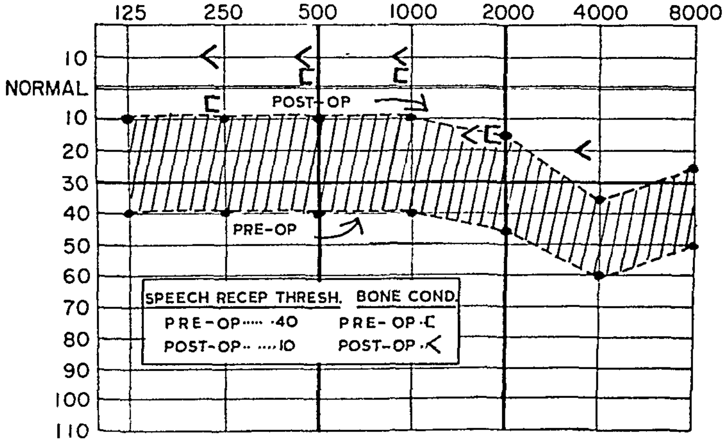


FIG. 3. R.C.

Audiogram of a 35-year-old woman 33 months after footplate mobilization directly at the footplate. A previous mobilization at the stapedial neck did not improve the hearing, and the crura were fractured. Direct footplate mobilization was performed 13 months later with this result, which has been maintained.

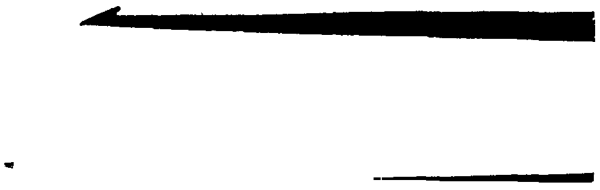


FIG 4.

Instruments used in making a fenestration through the footplate: the hooked oval window fenestrator and the sharp-pointed explorer.

the fenestrator is removed, the opening appears as a discrete hole through which one can see into a dark space—the vestibule. Usually a minimal amount of perilymph can be seen coming out of the fenestra. When a pledget of cotton is alternately pressed into the round window niche towards the round window membrane, the perilymph can be seen to rise and fall in the created fenestra.

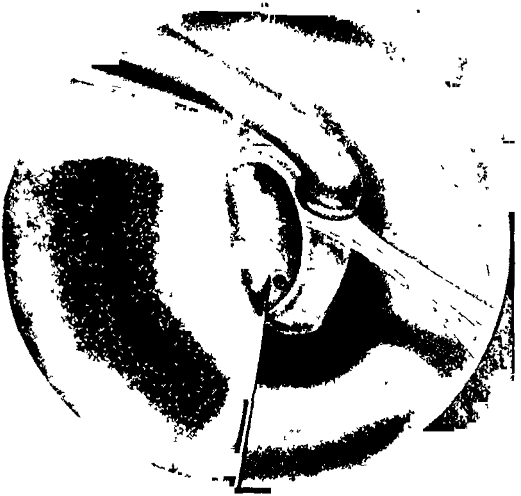
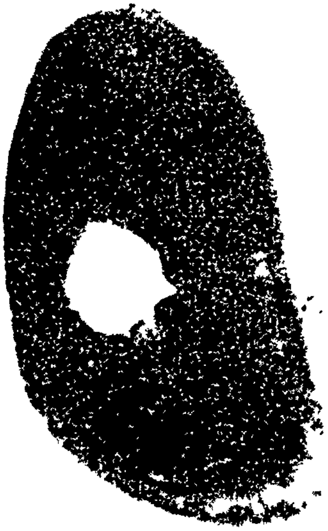


FIG 5

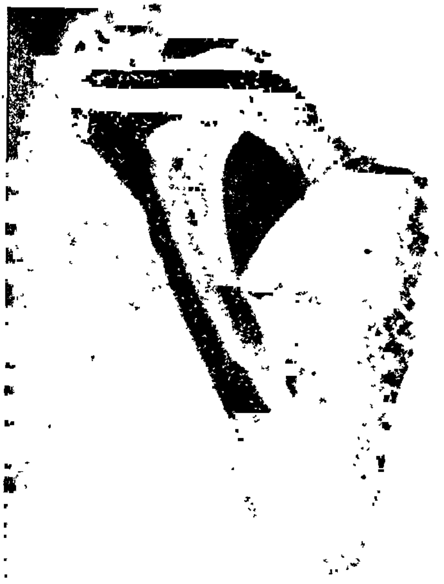
After all efforts at mobilization have been exhausted, fenestration of the oval window through the rigid footplate is employed The oval window fenestrator or the explorer is used at either of the sites shown

In order to achieve full visualization of the entire footplate for this delicate technique, it is sometimes necessary to remove the previously fractured crura and the incus as well (Fig. 6). Yet, despite this interruption of the ossicular chain, normal and near-normal hearing have been achieved by fenestration of the oval window. It should also be noted that in many cases of fenestration of the oval window, the footplate remains just as rigid even after the fenestra has been made. In other words, whether the ossicular chain is prevented from carrying out its sound transformer function by excision of the crura and incus, or by a still rigid footplate deprived of its piston-like action moving the cochlear fluid back and forth, hearing has been restored and *maintained* by creation of a fenestra through the footplate. The following cases illustrate this:

A.S. This 38-year-old man had bilateral progressive deafness for nearly five years. The pre-operative loss by pure tone and speech audiometry was 35 db in the right ear. In February, 1957, the right stapes was found to be extremely rigid at operation. The crura remained intact after repeated attempts to mobilize the footplate, but the incus was removed to obtain a fuller view of the footplate. A fenestra was made through the footplate which remained as rigid after the fenestra was made as before. The patient's hearing improved on the table and two weeks post-operatively, the hearing was at the 10 db level. Two months later the level was as it is in this audiogram, 5 above zero, which has been maintained for nine months to date. (Audiogram, A.S.; Fig. 7)



5A



5B

FIG. 5A.

Oval window fenestra created through the body of the stapedial footplate on a cadaver. The stapes was then removed from the specimen.

FIG. 5B.

Oval window fenestra created through the periphery of the stapedial footplate on a cadaver. The stapes was then removed from the specimen.

M.H. A 43-year-old woman had bilateral progressive deafness for 18 years and constant ringing tinnitus. Her pre-operative hearing loss by pure tone and speech audiometry was 65 db. She wore a hearing aid on the right side. In

August 1955, her right stapes was found to be very rigid and continued pressure against the neck finally caused both crura to fracture, leaving the rigid footplate A fenestra was made through the body of the footplate and the hearing improved on the operating table Post operatively, the hearing reached the 20 db level, which has been maintained for 27 months to date (Audiogram, M H, Fig 8)

C C A 41 year old woman had bilateral progressive deafness for seven years Her pre operative hearing loss was 40 db on the left ear and 35 db on

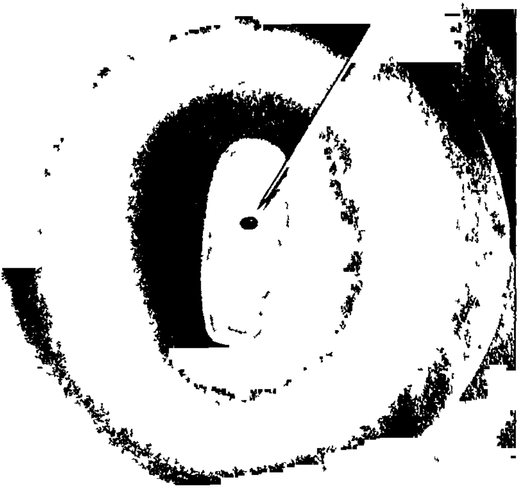


FIG 6

It is sometimes necessary to excise the previously fractured crura and the incus in order to obtain full visualization of the entire naked footplate before making the fenestra through the footplate at either of the sites shown

the right by pure tone and speech audiometry A successful mobilization at the stapedial neck was performed on the left ear in May 1954, and in December 1954, the same operation was performed on the right ear The hearing regressed in the right ear after eight months and in September, 1956 that ear was re operated The crura were found to have been fractured at the initial operation Fenestration was made through the body of the footplate with the hooked fenestrator The patient s hearing improved on the operating table and two weeks post operatively the speech reception threshold was at the 20 db level

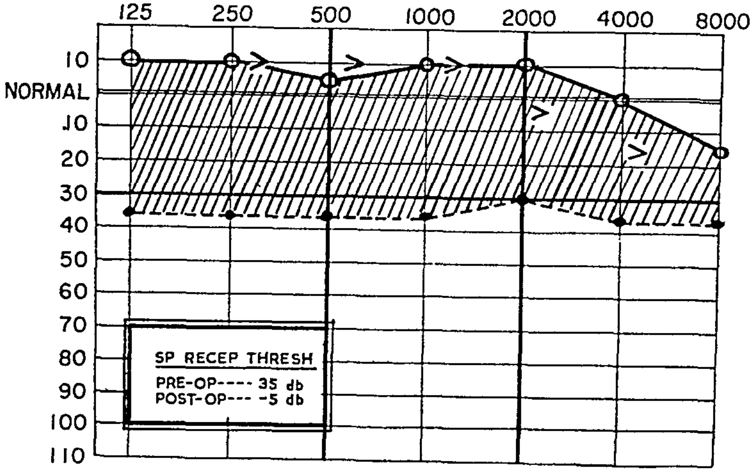


FIG 7 A S

Audiogram of a 38-year-old man with bilateral progressive deafness for 5 years, 9 months after a right fenestration of the oval window through the rigid footplate The incus was removed to obtain a better view of the footplate After the fenestra was made, the footplate was still as rigid as before The hearing improved on the operating table and two weeks post-operatively it was at the 10 db level A test two months later showed the hearing to have reached this level which has been maintained to date

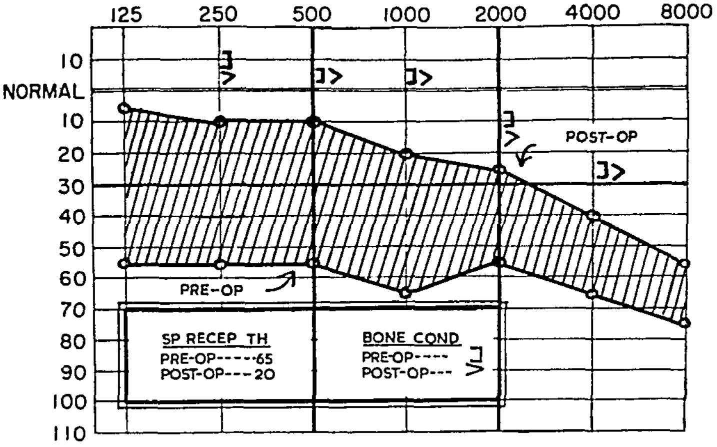


FIG 8 M H

This 43-year-old woman had bilateral progressive deafness for 18 years and wore a hearing aid on the right side In August, 1955, the right stapes was found to be extremely rigid and the crura were fractured during attempts to mobilize at the stapedial neck A fenestra was then made through the rigid footplate and the hearing reached the 20 db level post-operatively for pure tone and speech This improvement has been maintained as this audiogram shows, for 27 months to date from the pre-operative 35 db. The hearing continued to improve to the leve shown here, which has been maintained for 14 months to date. This patien has normal hearing on the right side following fenestration of the oval windov

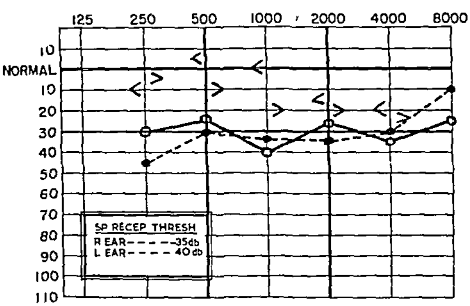


FIG 9 C C

Preoperative audiogram of a 41-year-old woman with bilateral progressive deafnes for 7 years

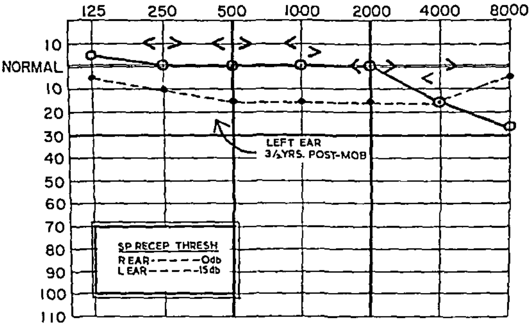


FIG 10 C C

Post-operative audiogram 14 months after fenestration of the oval window through the rigid footplate, on the right ear A previous mobilization at the stapedial neck had improved the hearing which then regressed following an upper respiratory infection The crura were found to have been fractured at the initial operation but the hearing improved after fenestration of the oval window to normal, as shown in this audiogram, and has been maintained for 14 months to date The left ear shows the hearing improvement maintained 3½ years after mobilization at the stapedial neck

and near-normal hearing on the left, 3½ years post-mobilization (Audiograms C.C., Figs. 9 and 10)

Restoration of hearing up to normal and near-normal levels by fenestration of the oval window has also been reported by Guillon of Paris at the Sixth International Congress of Otolaryngology in Washington (5). Dr. Michel Portmann of Bordeaux has also restored the hearing to normal by fenestration of the oval window even when the ossicular chain has been interrupted (6). In a personal communication, Altmann of New York has reported normal hearing, with closure of the air-bone gap, after removal of the fractured crura and incus, following which he fractured the footplate (7).

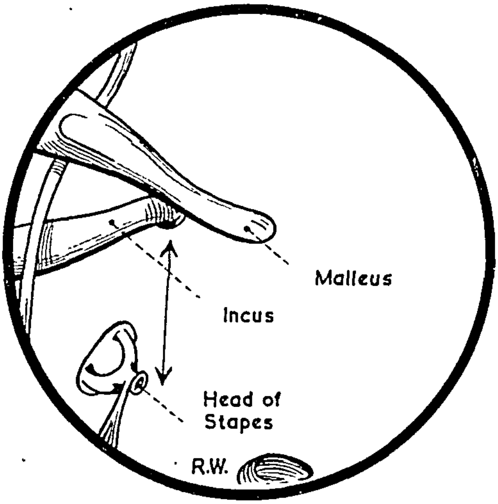


FIG. 11.

Schematic drawing of the wide and irreversible disruption of the incudo-stapedial articulation in an ear with normal hearing.

It would appear, then, from the experience gained so far in the development of stapes surgery from mobilization to fenestration of the oval window, that the Helmholtz concept of the transformer action of the intact ossicular chain is a *sufficient* but not a *necessary* condition for normal and near-normal hearing. This conclusion would seem to be justified by the next case to be illustrated. This case is an interesting one, which has given me much thought for several years. It is not a stapes mobilization and it illustrates the maintenance of normal hearing in a normally hearing patient after wide and irreducible separation of the incudo-stapedial articulation.

I.O. A woman, 29 years of age, suffered for several years from severe attacks of vertigo, which were diagnosed as bilateral Ménière's disease. The hearing on repeated tests was always found to be normal in both ears. On March 17th, 1954, a bilateral chorda tympani nerve section was performed through the external auditory canal in order to relieve the vertigo (8). No bone of the posterior canal wall was removed and only the very edge of the incudo-stapedial junction could be seen During the operation on the right ear the long process of the incus was inadvertently hooked outward for a short distance, along with the chorda tympani nerve The dislocation of the stapedial end of the long process was so wide that it almost touched the middle of the hammer handle (Fig 11) *The incus became rigidly locked in the incudo malleolar articulation* as if in a vice, and could not be moved from this abnormal position The disconnected articular surfaces of the incus and stapes were separated by at least 6 mm The drum was replaced

In the 3½ years since the operation this abnormal position of the long process of the incus has not changed and can still be seen through the normal right drum *The drum is in normal position, is not scarred or retracted by any* adhesions and is not in contact with the head of the stapes Repeated audiological tests since the operation show the normal hearing as before the operation Since section of both chorda tympani nerves, incidentally, all attacks of vertigo in this patient have disappeared (Audiogram, I O Fig 12)

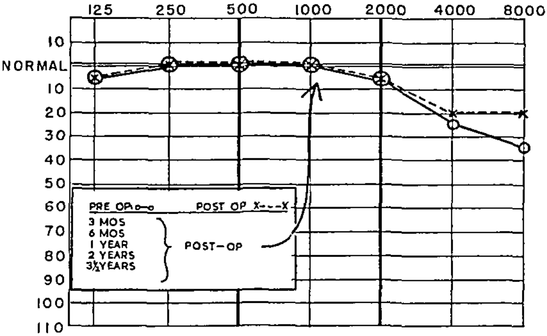


Fig 12 I O

Pre and post operative audiogram of a 29 year old woman with Menière s disease Normal hearing has been maintained for 3½ years after wide and irreversible separation of the incudo stapedial articulation in the right ear during section of the chorda tympani nerve

If normal hearing can be maintained for so long a period after such a disruption of the ossicular chain, it would at least make intelligible the normal and near-normal hearing after fenestration of the oval window, where the ossicular chain is interrupted and/or no longer functioning There appears to be, therefore, structural conditions other than the intact transformer mechanism described by Helmholtz, which are capable of producing normal hearing These various conditions may be regarded only as known parts of a total phenomenon, the nature of which is not yet completely understood Problems such as these remain to be solved theoretically as well as practically

One of the most puzzling of these problems is related to the fenestration through the footplate. I observed early in this technique, as others have since, that in some cases the marked improvement occurring immediately after the fenestra was made, disappeared as soon as the drum was replaced. However, a myringotomy performed immediately on the table restores the hearing to that improved level. Why does this happen in some cases and not in others? This should be a stimulating problem for the acoustic physiologists.

While we search for the answers to these problems, it is, however, possible after more than five years, to evaluate the advantages of stapes surgery for the relief of otosclerotic deafness. For the patient, these advantages are many: an overnight stay in the hospital, quick resumption of normal activities; no post-operative treatment or crippling vertigo. (After fenestration of the oval window there may be slight or uncommonly severe vertigo persisting for only a few days.) In experienced hands the danger of complications is negligible: in over 3,000 cases of stapes mobilization and fenestration of the oval window I can report no instance of total deafness, facial nerve paralysis or any other significant complication.

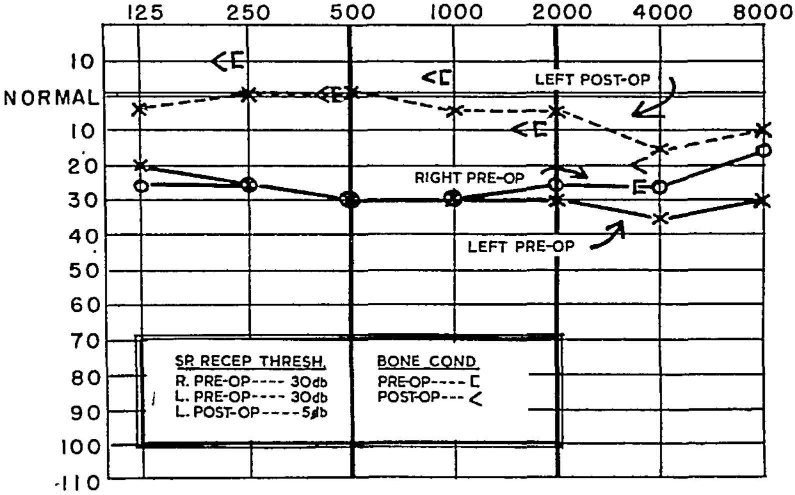


FIG. 13. E.W.

Audiogram 30 months after mobilization at the stapedial neck on the left ear in a patient with bilateral pre-operative loss of 30 db by pure tone and speech audiometry.

The audiological basis of case selection for the surgery of otosclerosis is the relationship between the pure tone thresholds of hearing by air conduction and by bone conduction, the so-called air-bone gap. A normal or near-normal threshold by bone conduction accompanied by a significant loss by air conduction is considered to be the most suitable condition for stapes surgery. When the air-bone gap is eliminated in these cases, by stapes mobilization or fenestration of the oval window, normal or near-normal hearing results

Any significant air-bone difference (anything more than 15 db in the speech range) indicates a possible case for stapes surgery, because any one of the three procedures described here can restore all of the hearing within the cochlear potential—that is, up to the bone conduction level No other surgery for otosclerotic deafness has been able to achieve this Losses as little as 25-30 db and as great as 95 db by speech reception have yielded to this type of surgery for gains in hearing of 15 to 45 db The following two cases illustrate (Audiogram E W , Fig 13 Audiograms, C W , Figs 14 and 15)

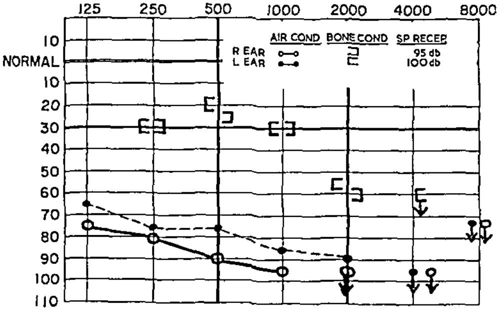


Fig 14 C W

Pre operative audiogram (right ear) of a profoundly deafened 45 year old woman Mobilization was performed to permit successful use of the hearing aid

In the case of C W , several things are noteworthy She had a severe hearing loss bilaterally, with markedly depressed bone conduction Her discrimination scores of 50 per cent in the right ear and 62 per cent in the left were taken at levels of 10 and 15 db above her speech reception thresholds at the 110 db level, the limit of the test equipment

Despite the profound hearing loss and poor bone conduction, the relatively high discrimination scores obtained at such low sensation levels for the patient, suggested a good cochlear reserve The substantial air bone gap also entered critically into the decision to operate Following mobilization of the left rigid stapes, the average pure tone loss rose in that ear from 83 db to 68 db and the speech reception threshold from 95 db to 75 db The discrimination score on the operated ear was tested post operatively at the same audiometric level as in the pre operative test 110 db on the audiometer This was now 35 db above her new speech

It might be relevant at this time to repeat a word of warning included in my report to the American Triological Society meeting in Florida in 1955 (10):

> "It is my conviction that procedures of this kind should be performed only by those skilled in endaural surgery . . . Even the most skilful surgeon should perform the operation on at least 50 fresh cadavers before the first patient is operated on . . . because of the great number of anatomical variations.

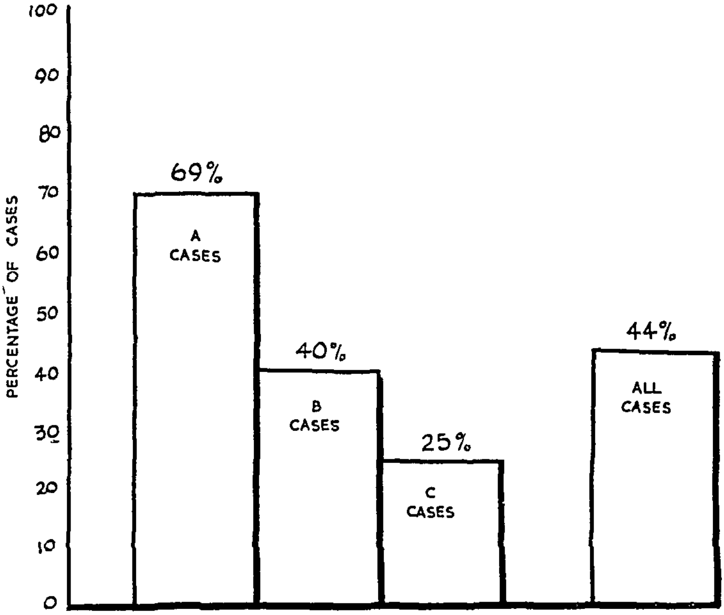


FIG. 17.

Restoration of hearing to the 30 db level or better in A, B and C cases following fenestration of the oval window (136 cases)

> "This operation appears deceptively simple, yet learning to perform it skilfully is not . . . to some this is an unfamiliar approach to intratympanic surgery.
>
> "Personal communications to me of some of the early results of the operation performed by others on the living . . . indicate insufficient practice on the cadaver."

With regard to fenestration of the oval window, I should like to repeat here what I said at the Sixth International Congress of Otolaryngology in Washington (11):

> "The physical encounter or contact or collision of the point of the instrument with the footplate must first be *felt* unmistakably before penetration is made, no matter how well one 'sees' the contact of the

instrument with the footplate Also, the point of the instrument must not only be seen to penetrate the footplate into the vestibule, but it must be felt to penetrate it for not more than a millimeter The tactile and kinesthetic sensations have been found to be more reliable than the visual, although the associated sight and feel are desirable '

Statistical results of stapes mobilization at the neck and footplate have been widely reported by myself and others The first report on the statistical results of fenestration of the oval window was made last May

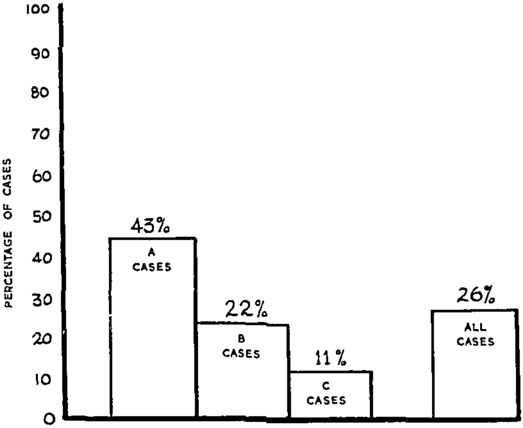


FIG 18

Per cent of cases of fenestration of the oval window which reached the level of 20 db or better by pure tone and speech audiometry) 136 cases)

in Washington at the Sixth International Congress and I should like to restate them here (10)

Of 136 consecutive cases, 72 per cent of the A cases, 55 per cent of the B cases and 43 per cent of the C cases showed significant improvement in hearing (15 db or more gain over the pre-operative level) Combining the results in all three categories, thereby including the least suitable with the most suitable, 57 per cent showed such improvement (Fig 16)

The following figures show the restoration of hearing to the 30 db level or better 69 per cent of the A cases, 40 per cent of the B cases and 25 per cent of the C cases reached these levels, while the combined figure for all three categories is 44 per cent (Fig 17)

The percentage of those whose hearing improved to the 20 db level or better are: 43 per cent. of the A cases, 22 per cent. of the B cases and even 11 per cent. of the least suitable C category, for a combined total of all cases—from the most to the least suitable—of 26 per cent. Better than one out of every four cases of varying suitability achieved restoration of hearing to the 20 db level or better by fenestration of the oval window through the rigid footplate (Fig. 18).

The ultimate goal of any surgery for otosclerotic deafness, including the classical fenestra nov-ovalis, is of course, the restoration of maximum possible hearing in both ears. In view of the effectiveness of these three surgical procedures, as described here, it is suggested that the otosclerotic patient be advised to look upon stapes surgery as the first step, to be repeated, if necessary, until this goal has been achieved.

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STAPES MOBILIZATION

By TERENCE CAWTHORNE (London)

FOLLOWING the example of Dr Rosen, many surgeons in different countries have found that in otosclerosis it is often possible to improve the hearing by breaking through the bony adhesions binding the footplate of the stapes, usually at its anterior end, to the adjacent rim of the oval window

Why this bony bond does not usually, or even often, re-unite and the deafness return has been a pleasant surprise, and in many circumstances stapes mobilization is now regarded as the operation of first choice in the surgical treatment of otosclerosis

It has the great attraction from the patient's viewpoint of being a slight, one might almost say trivial, operation It is but rarely followed by dizziness or by infection and if it fails to improve the hearing, a fenestration operation still has the usual chance of succeeding

On the other hand it does not as yet offer as good a chance of restoring hearing as the fenestration operation, and an initial improvement in hearing is less likely to be maintained after mobilization than after fenestration, though it is possible to restore hearing again by re mobilization

Before giving you my own experience, such as it is with this delicate and interesting operation, I would like to offer a few observations on the stapes itself Besides being the smallest bone in the body it is also the most fragile, and it does not need much force to break through either the neck, the crura, and, in particular, the slender anterior crus, or through the footplate

I have taken the opportunity of studying and manipulating the normal and unimpeded stapes in the living in the course of removing it so as to reach and remove the membranous labyrinth *viá* the oval window and to destroy cochleo-vestibular function disordered by Meniere's disease Afterwards the oval window is closed by replacing the intact stapes To do this without damaging the stapes has made me appreciate the meaning of the phrase "Handle with care" As, however, the stapes moves freely to the touch it is not so easily damaged as when it is partly fixed by otosclerotic bone It is interesting to see quite a wide space in front where the anterior crus is quite free of the rim of the oval window and the anterior edge of the footplate itself can be seen through the annular ligament when slight backward pressure is made on the neck

In otosclerosis, however, the appearance at the anterior end of the oval window is usually quite different. The anterior end of the oval window is narrowed so that its lips appear to embrace the anterior crus and there is no free space at the depth of which the anterior margin of the footplate can be seen.

I should say, of course, that what I have just described can best be seen using the Zeiss microscope with 16 diameters of magnification.

Furthermore, the stapes in otosclerosis can become set so to speak at different angles, but in most of the cases I have studied so far the main, if not the only, bony bond between the oval window and the stapes seems to have been at the anterior end and it is at this point that the mobilizing force should first be applied in order to loosen the footplate.

I think that all who have tried mobilizing the stapes started by applying traction on the neck in the way so ably described by Dr. Rosen. As he has repeatedly said, not only the amount of force but the direction of the traction as well needs to be watched in order to avoid fracture of the crus, the crura or the neck of the stapes. If then traction is applied properly it is a tug-of-war between the otosclerotic bone and the ability of the stapes to stand up to traction. Of course the stronger always wins, sometimes it is the otosclerotic bone which gives way, sometimes the stapes. Another and possibly safer way is to push on the head of the stapes, the pressure usually being applied through the incus. Here again the stapes may give way before the bony adhesion.

Fowler deliberately cuts through the anterior crus and cracks across the footplate, thus separating the fixed from the mobile part of the bone. That it works I know because I have done it accidentally and the patient eventually heard better; though I must confess that I was apprehensive about the result particularly as the patient was very dizzy indeed for some days. I still would hesitate to do this deliberately.

Heerman uses very fine gouges and chisels the bone free. I have seen Schuknecht do this and have discussed it with him and he believes it to be a good method. Certainly I think that it is likely to be useful in advanced cases where the footplate is more likely to be firmly fixed.

My own preference is to try and mobilize at the level of the footplate using Rosen's curved needle and trying not to touch the footplate or the crura except at the very beginning and the end when I test the mobility of the stapes by the lightest pressure on the incus. In this way it is usually possible to avoid damaging the stapes and the proportion of cases showing significant improvement in hearing has increased from 45% when working at the neck to 72% when working at the level of the footplate.

Against this I must say that there is a slight but definite increase in the tendency to relapse the more one actually works on the otosclerotic bone. For that reason I am trying to work down just in front of and on either side of the anterior crus rather than through the bone in front of the anterior crus.

There is usually no doubt when the stapes has been successfully mobilized. It bobs up and down like a cork in water in response to the lightest touch on the long process of the incus. It is often helpful to check the hearing for pure tones at the time of operation; though there is usually no doubt when the hearing has been properly restored.

If the stapes can be mobilized without actually touching it either by applying pressure through the incus in early cases or by pressure with the *needle or curved seeker near where the anterior crus joins the footplate* the chance of damaging the stapes is reduced to a minimum. Sometimes, in addition to pressure at the anterior end, it is also necessary to apply pressure near where the posterior crus joins the footplate before there is a satisfactory hearing improvement.

I *find it helpful to use the microscope for all stages of the operation,* working at 10 diameters of magnification for the incision and reflection of the flap and removal of the bony meatal margin which I do with fine and shortened Jenkins gouges. Once the middle ear has been opened and the ossicular chain exposed the magnification is increased to 16 times.

Hearing is checked with a pure tone audiometer and the flap back in position; this may have to be done two or three times before the operation is finished. No case is regarded as improved unless there is more than an average of 10 decibels improvement over the pre-operative hearing for pure *tones between 500 and 2,000 c.p.s.*

I have done 25 fenestrations after unsuccessful mobilizations and 22 have been successful. Seven cases have been remobilized with a satisfactory improvement in five.

These results encourage me to believe that stapes mobilization is an important advance in the surgical treatment of otosclerosis.

STAPES MOBILIZATION

By R. S. VENTERS (Carlisle)

THE invitation to present this paper in association with Dr. Rosen and Mr. Cawthorne was indeed a surprise, and I feel much honoured.

Though I admit, that at first I was a sceptic, the impact of Dr. Rosen's original article was profound, for if mobilization of the stapes was, in my opinion, a questionable procedure, the feasibility of opening into and viewing the tympanic cavity with impunity was not. I confess to having done no preparatory cadaver surgery and, though this may shock some, I would say in mitigation of my crime, and more especially to the younger members, that I have had more than twenty-five years of daily familiarity with the temporal bone. This is what happened in Case No. 1. My enthusiasm was unlimited and I set myself the task of perfecting the technique and laid plans for a carefully recorded series of cases.

Though Dr. Rosen's original description of the operation cannot be bettered, there are, having worked out the operation for myself, several points in technique that are worthy of some emphasis. I prefer a general anæsthetic in order to ensure not only freedom of movement, but freedom of speech. An immediate audiograph is of questionable value, for the sedated patient has a reduced cochlear response and the non-sedated a euphoric inaccuracy. The degree of mobilization bears no relationship to the ultimate hearing gain and in any case I do not like my theatre cluttered up with more gear than is necessary. The first 30 cases were operated on with spectacles alone. Since the acquisition of a Zeiss operating microscope, however, I have found technically that it is easier and quicker to proceed with spectacles to the point of full reflection of the meatal flap and tympanic membrane. The subsequent inspection and mobilization is done with the aid of the microscope.

The limited magnification, and in consequence the greater depth of focus of the spectacles, allows one to keep in view the whole of the tympanic membrane and the deep half of the canal without fiddling around with the speculum. The edge of the tympanic membrane is approached first just below the tympanic notch, and the bluey grey of the intratympanic mucosa must be well defined before entering the cavity and reflecting fully the tympanic membrane from the sulcus. In eight out of ten cases the chorda tympani nerve remains tucked close to the tympanic bony ring and must be recognized. It is readily detached from its mucosal bed with a fine pointed seeker and pushed medially. With increased experience I found that it was never necessary to sacrifice the nerve.

With the exception of two cases, apart from some sundry small adhesions, I found the cavity to appear perfectly normal. One showed quite clearly fusion of the crura and in another the cavity contained glutinous mucus which would not even enter the sucker and had to be removed like winding up a skein of wool. I presume a case of faulty diagnosis.

The high-powered view of the tympanic cavity fascinates me and I admire greatly the descriptive drawings of Rosen, Scheer, House and others, depicting with great vividness and skill the footplate and crura, but I cannot relate what I see to what they seem to see and draw. Even with removal of part of the bony tympanic ring the fenestra-ovalis with its stapedial footplate remains to me terra incognita in a high percentage of cases. The long process of the incus, the posterior crus and the apparent depth of the fenestra with a frequent overhanging upper rim presents quite a problem to my visual field and manipulative dexterity. There is also the overall mucosal blanket and above, the facial nerve. In some, anatomical variation does allow to a view of the lower and anterior portion of the fenestra and in an attempt to visualize the annulus, I have reflected the mucosal blanket, but its vascularity is such that hæmorrhage can be troublesome and difficult to control.

I have observed, however, and in spite of the visual limitation, that in several there seemed to be no line of demarcation between the footplate and the fenestra. In most the footplate itself seems to be so thin as to be almost transparent. I had the impression of almost looking into the vestibule and an apt description would be thin ice on a pond. On touching this lightly with a fine probe, the plate perforated with limited radiating multiple fractures. Direct, visually controlled mobilization or fenestration of the footplate seems to me to be applicable only in a small percentage of cases.

In recording operative details it was soon apparent that there was one finding almost common to all, fixation of the footplate. The stapes was mobile in only 18 of my series of 130 ears. As this must of necessity be a personal assessment, I took considerable care in determining accurately, whether or not fixation was complete. There seemed no doubt about the fixation in almost every case, but there was a very distinct varying difference in the tactile sense of solidity. Some seemed almost to be monolithic and others gave the sensation of being fixed to a less secure base. At ten magnification and over, this tactile sensation of insecurity of the latter could be related to visual movement and along both the long and short axis.

It is my opinion that this movement is the result of distortion or bending of the footplate rather than even limited mobility of the annular ligament.

It would seem that the long-accepted clinical evaluation of total stapedial fixation has been well confirmed both visually and by direct tactile assessment. The audiograms do not seem to differ and in no way indicate the known varying pathological lesions either as to extent or position, healed or active. I believe that I can now assess this subtle degree of fixation with a fair measure of accuracy and can determine immediately those cases likely to be successful and those likely to fail. With increasing experience I have broken fewer crura and now desist should my tactile sense of solidity be such that crural fracture is likely and I fenestrate at a later date.

What is more important, is the fact that I now achieve a much higher percentage of successful results and I attribute this not only to a better selection of cases, but to a more accurate application of pressure on the right place and in the right direction. The mobilizer must be placed at the neck and the pressure applied posteriorly, slightly downwards, and in the long axis of the footplate.

The procedure I follow does not vary in any way from that described by Dr. Rosen, but I have three observations to make that to me are important.

It is not essential to observe a light reflex from the round window to assure oneself of a successful outcome. Various factors are involved such as the direction of the primary light, depth of the membrane and amount of reflecting fluid in the niche. There is one other factor, however, that could be overlooked; there may be insufficient movement at the footplate to produce a fluid wave in the perilymphs for visual appreciation of round window movement by light reflex.

Another observation concerns the stapedial tendon. Normally the stapedial tendon has the appearance of being taut and straight and, on application of a probe to its surface, one can appreciate a certain amount of bounce. On successful mobilization it will be seen that the tension has gone and it may even have a slightly bent appearance and bounce is replaced by sag. I deduce from this that the crural arch is displaced posteriorly and downwards and does not return to its previous normal position.

Thirdly, in no case have I observed either the presence of blood or leakage of perilymph around the footplate area and assume that the mucosa remains intact.

I shall have more to say about these observations later.

Table I. My series consists of 108 patients, 130 ears and all operated on under the National Health Service. Twenty-two patients had both sides operated on. All had the poorer ear operated on first.

To begin with, I accepted for operation with little discrimination, almost all cases of clinical otosclerosis that came my way, but soon adhered largely to the Shambaugh-Carhart formula and classification as for fenestration. It would seem to me, and put simply, that the flatter the audiograms and the greater the air/bone gap the more likely a successful result. However, there were so many exceptions to this that *one would be bold* *indeed to build up a patient s hope on the presentation of an ideal audiogram or refuse operation on the evidence of a less promising one*

On the presentation to the patient of a truthful statement as to the considered chances of success I have experienced no difficulty in persuading patients to have the operation It may be that in my more northern climate the patients are more phlegmatic, but more important is the fact that the bonfire telegraph of the Lakeland and Scottish Border hills reminiscent of the depredations of the Scots and English, still exists and they clamour to have the operation done

TABLE I

STAPEDIAL MOBILIZATIONS
(OCTOBER 1955 OCTOBER 1957)

	Males	Females	Total
No of patients	50	58	108
No of patients—single operation	36	50	86
No of patients—double operation	14	8	22
Total no of ears	64	66	130

Table II This Table shows the immediate results and you note the very successful percentage relative to the moderately successful No audiograph is required to persuade one that a case has been very successful The patient knows, everyone directly and indirectly associated with the ward knows and emotional upsets and tears are not uncommon At times it has been quite embarrassing Mothers have had to be parted from their children in order to undergo a period of acoustic rehabilitation and others have kept indoors to avoid traffic noise

TABLE II

IMMEDIATE POST OPERATIVE RESULTS ON 130 EARS
(OCTOBER 1955 OCTOBER 1957)

	Number	Percentage
Very successful	71	55
Moderately successful	20	15
Failures	39	30

Table III My first cases were done in October, 1955, and I have followed up all cases done prior to June 1st, 1957, either by personal examination or by means of a questionnaire The percentage of immediate successful results is 70 per cent and the percentage of successful results maintained out of the total to that date is 53 per cent

TABLE III

Number of Ears Successfully Operated on Prior to June, 1957

	Number	Percentage
Number of initial successes: Very Successful and Moderate ..	73	70
Number of successes maintained: Very Successful and Moderate ..	55	53

With the exception of one case with a transient middle-ear infection, there have been virtually no complications. No patient has experienced vertigo and in about 50 per cent. of all cases with pre-operative tinnitus, the tinnitus disappeared.

Though the percentage of very successful maintained results, even with a more critical criteria of acceptance and increased skill, may not greatly exceed 60 per cent., *the social rehabilitation of those as the result of a safe and, to the patient, a minor operation, cannot be ignored by any practising otologist.*

When I met Dr. Rosen I bombarded him with questions and his invariable modest reply was "I do not know" and my reading of the literature would lead me to believe that there are still many questions unanswered. It is insufficient to state that the "proof of the pudding is in the eating of it". I frankly do not believe, except perhaps in a very small percentage of cases, that true stapedial mobilization, or if you like stapediolysis takes place.

I find it equally hard to accept that it is justifiable to destroy permanently the ossicular chain, in order to knock a hole in the footplate or to bite a piece out of its edge. Surely these holes must be covered eventually, if not by bone, by dense scar tissue. Direct attack on the edge of the footplate is more logical if you can see it and define it without being destructive, but early in this paper I confessed that I cannot define the footplate with any degree of accuracy, except in a small percentage of cases. Again, *if that something which happens, on pressure being applied to neck, does not happen*, there must be a fairly extensive fusion between the rim of the fenestra and the plate. I grant that this fusion may be broken by direct visual application of elevators, but the resultant contiguous, rough, saw-like, bony edges can hardly be conducive to free movement and the maintenance of mobility.

Though the numbers are small I certainly got the impression that if the operation was successful on one side there was a good chance of success on the other. Twenty-two patients had both ears operated on and a double successful result was achieved on fifteen. I infer from this, that if the pathological lesion, both as to site and extent, has been such as to ensure a successful result on one side, it is reasonable to suppose that a similar lesion exists on the other. Patients who have had a successful result require no persuasion, indeed are anxious, to have the other side done, and I am chary about advising further operation in the unsuccessful.

I do not ponder on why the successful maintained results do not much exceed 50 per cent., for the varying pathological lesions must prevent a successful outcome in any given number of cases. This operation can only be part of, but must be part of, one's armament in the treatment of otosclerotic deafness. I wonder what really happens at the footplate? With a normal cochlear response and clinical fixation of the footplate, restoration of a freely-moveable ossicular chain should produce normal hearing, but that does not happen, and why not? Why is the hearing maintained at all and why does tinnitus disappear in some and not in others?

Just as the very successful hearing result leaves no shadow of doubt that it is successful, I believe that a successful operation is something just as well defined, at least to the experienced operator. On the application of varying pressure the crura rotate backwards and downwards, with or without an audible click, in a most definite fashion. There is no sense of a sudden collapse as occurs with broken crura, but rather a controlled sensation of sinking backwards and downwards. When this occurs I feel reasonably certain that the result will be good.

My observations and results do not, I think, materially differ from those of others employing the same technique, and though there are imponderables there are certain well-established and definite findings. These can be summed up as follows:

(1) Clinical fixation confirmed at operation;
(2) A successful operation with consequent good results;
(3) Maintenance of improved hearing in at least 50 per cent. of cases.

Fig. 1 shows diagrammatically the stapes with direct lines of stress. The thickened edge of the footplate is substantially held to the rim of the fenestra by the annular ligament, and it is well known that on forcible removal of the stapes both crura and footplate will often fracture before the ligament gives. The upward arrows indicate the thrust of the perilymph in the vestibule, which for practical purposes can be looked upon as a closed cavity.

Fig. 2 represents what I think happens. The stress constructional engineers refer to this as "punch and sheer". The posterior crus, the stronger of the two, is pushed backwards and downwards, and, especially in cases where the plate is thin, a variable portion of the actual plate is carried into the vestibule. There must of necessity be a break anteriorly, and as no break of the anterior crus can be felt I assume that the fracture, in most cases, takes place near the plate. The closely adherent unbroken mucosal blanket remains attached to the fragment and it is likely that this forms a false ligament.

Fig. 3. This is similar except that it attempts to show the situation where the otosclerotic lesion invades the anterior part of the plate and crus.

I realize that much of this is based on conjecture, but it would seem to explain *that something* which happens. It would account for the permanence of the successful result, and it might even explain why hearing is not restored to nearer normal due to the reduction in force and amplitude of the fragment relative to complete stapes.

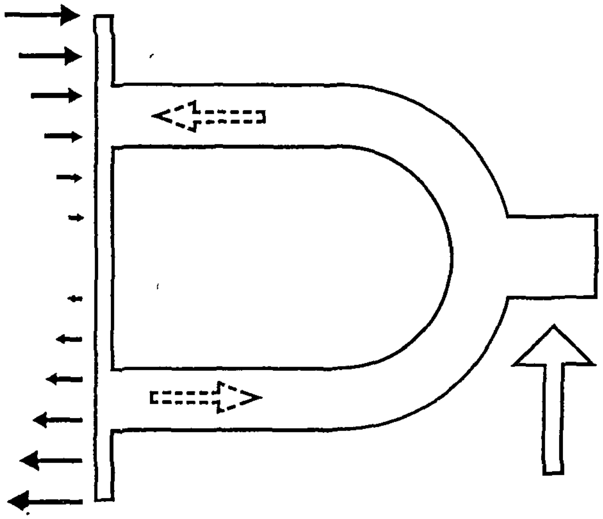

Fig. 1.

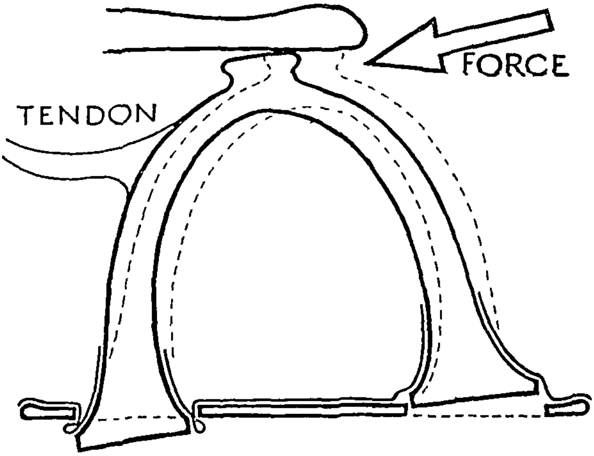

Fig. 2.

In conclusion

The operation as described by Dr Rosen, is an established effective procedure in the relief of deafness due to otosclerosis

The operation is not easy and demands a high degree of manipulative dexterity and tactile sensibility

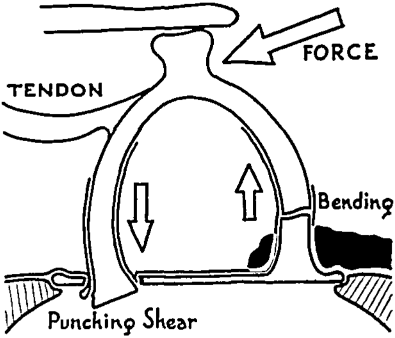

Fig 3

SUBMUCOSAL DIATHERMY OF THE INFERIOR TURBINATES

By J. F. SIMPSON and J. GROVES (London)

In this article we describe a method of treating nasal obstruction due to engorgement of the inferior turbinates by means of unipolar submucosal diathermy. The results of treatment in a series of a hundred cases are given. These cases represent a large and often depressing section of rhinological practice. They fill the surgeries and clinics, and their consumption of "drops" is a mainstay of the pharmaceutical industry.

Chronic turbinal engorgement, when due to sinus infection or specific nasal hypersensitivity (true allergic rhinitis) is often relieved by treatment of the underlying condition. Still there remain many in whom such causes are not apparent, or in whom appropriate treatment fails to relieve the nasal obstruction. Symptomatic treatment is then necessary, and linear cauterization of the inferior turbinate with the galvano-cautery is often successful. By the formation of scar tissue adherent to the turbinal bone excessive swelling is controlled. This minor out-patient procedure has the disadvantage of destroying all the mucosal epithelium which lies in the path of the hot wire. During healing there is the discomfort of crust formation, and the risk of adhesion formation between the septum and the raw surface of the turbinate.

Historical

In the past fifty years several techniques have been described which do not have these disadvantages.

Neres (1907) buried a gold needle in the turbinate by running it along the medial surface of the bone. A measured galvanic current was applied for several seconds. In developing this technique Neres recognized the advantages of not damaging the mucosal surface.

Horn (1908) described a submucosal technique in which he dissected the mucosa from the medial surface of the turbinal bone through a small anterior incision. A hot wire electric cautery was then passed along inside this mucosal pocket, to coagulate the submucous layers without damaging the surface.

When high frequency diathermy currents became available they were used in at least two different ways. Bourgeois and Poyet (1922), Castex (1929), Sinskey (1932), and Jaros (1933) described techniques in which diathermy current was applied by various types of electrode to the *surface* of the nasal mucosa. The effects aimed at varied from mild warming of the tissues over a wide area, to actual coagulation in a linear application to the inferior turbinate. Good results were claimed, although indications for treatment varied widely, from the common cold to "nasal dysmenorrhœa".

The diathermy current was also used to produce a *submucous* linear burn, such techniques being logical developments of the methods of Neres and of Horn. Beck (1930) gave an account of his method. A unipolar electrode was improvised, using a Hagedorn needle which was insulated, except for 2 mm. at its tip. Through a puncture in the anterior end of the turbinate this electrode was pushed backwards between the bone and the mucosa for the full length of the turbinate. The needle was then slowly withdrawn while a heavy coagulating current was applied. Two or more "runs" were made according to the amount of shrinkage required. Beck had treated over fifty cases, but his follow-up period had been short and his results were not given in full detail. Nevertheless, his patients obtained relief from nasal obstruction in nearly every case. A few of them needed a second treatment.

Hurd (1931) and Morrison (1931) each described bipolar electrodes. Two wires, about 4 cm. long and parallel to one another were buried in the length of the turbinate through anterior punctures. Coagulating current was passed, which acted simultaneously on all the tissues embraced between the wires. So far as the literature is concerned there the matter rested until Harris (1945) in a short article, described the use of an ordinary, uninsulated straight needle as a unipolar electrode.

In 1948 Richardson, in what is perhaps the raciest contribution on this subject, gave an enjoyable account of the treatment of a hundred and seventy-five cases by submucosal diathermy. He used a bipolar electrode like Hurd's, and only six patients failed to obtain relief. The most lasting success was treated eight years before publication. Eight patients returned for further treatment within two years.

Shahinian (1953) published a very full account of the bipolar method, describing his own modification of Hurd's instrument. He treated four hundred and twelve cases, some with success lasting up to twelve years. Six with severe allergy needed further treatment after two years.

The results obtained by Shahinian and by Richardson are impressive and it is curious that the literature on this subject is so scanty. Shahinian suggests that the original technique described by Hurd was so satisfactory that no other comment seemed to be needed. If this were indeed the case one would expect to read some account of submucosal diathermy in our standard textbooks, but we have failed to find any description.

Electrodes and Technique

The advantages of the bipolar electrode are described by Shahinian who points out that it results in a flat strip of "cooked" tissues, whilst the unipolar electrode can only produce a line or cylinder of coagulation. On the other hand, if the size of the turbinate requires it, there is no difficulty in making several parallel "runs" with the unipolar electrode, and the single wire is easier to control and position.

At St. Mary's Hospital a unipolar electrode has been in use for some years. The model illustrated here (Fig. 1) was made by Messrs Matburn, Ltd., of Red Lion Street, London, W.C.1, and can best be described as a rather large myringotome. The shank is encased in a layer of insulating material from the handle right up to the base of the triangular blade. An indifferent electrode is strapped to the patient's leg, and the knife point is pushed into the most prominent part of the anterior end of the inferior turbinate. It is then pressed backwards deep to the mucosa, keeping close to the medial surface of the bone. It is important to follow with the tip of the electrode any tendency of the turbinate to deviate laterally in the deeper part of the nose, otherwise the mucosa may be "button-holed". When it is judged that the posterior end of the turbinate has been reached (Fig. 2) the circuit is closed and the electrode is withdrawn, taking some ten to twenty seconds to traverse the full length of the turbinate. The rate of withdrawal of the electrode must be slow

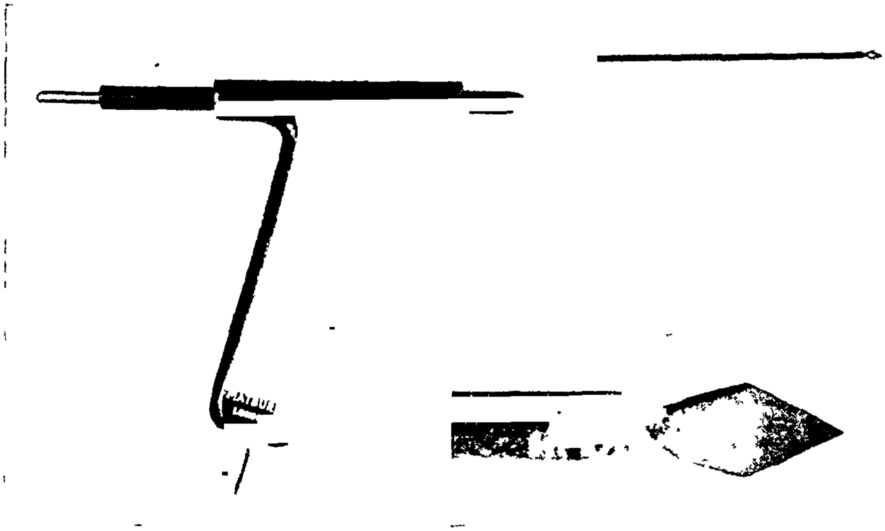

FIG 1

The unipolar electrode Note the thumbscrew, which fixes the removable electrode in the handle The inset is a magnified view of the tip of the instrument enough to allow coagulation, but fast enough to avoid charring If the electrode does not glide smoothly, intermittent bursts of current should be applied, the flow being interrupted while the electrode is moved in steps of a few millimetres at a time A linear submucosal burn results without apparent injury to the mucosal surface

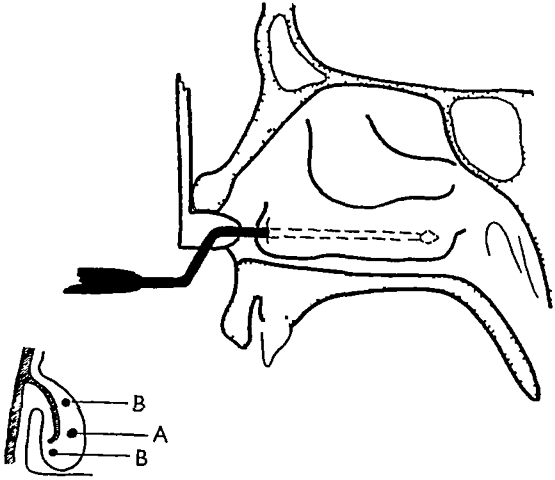

FIG 2

Diagram showing the electrode in position prior to coagulation during withdrawal The inset illustrates

(*a*) the usual line of coagulation
(*b*) additional lines in which a second or third run is sometimes necessary

The Coagulating Current

Most diathermy machines have no indication of the current flowing or the frequency employed Shahinian and other authors have used raw steak in which to calibrate their instruments, using a current which produces visible coagulation without charring in an arbitrary time measured in seconds These authors were using bipolar electrodes

With a unipolar electrode and a moving point of coagulation the variable factors are too numerous for this type of calculation In our cases we have used an arbitrary setting of the current at which coagulation is seen by the operator as a blanching of the mucosa over the electrode

(This blanching is only clearly seen when the turbinate has not been shrunk by topical anæsthetic solutions—general anæsthesia was employed during these trials.) Readings taken with a thermocouple type ammeter connected in the lead to the electrode have shown this current to lie between 0·4 and 0·5 amps. The diathermy machine was of the spark-gap variety with a fundamental frequency of 100 Kc./sec. and an infinite number of harmonics.

Anæsthesia

Children and nervous adults have been treated under general anæsthetic. Intubation and a pharyngeal pack, or a cuffed endo-tracheal tube, must be used, because occasionally brisk bleeding can occur from the site of turbinal puncture.

Local analgesia has proved most successful for this technique and allows it to be used as an out-patient procedure. With local, as compared with general anæsthetic, the freedom from bleeding has been striking. The nose is first sprayed with cocaine or xylocaine solution and an injection is then made of 0·5 ml. of xylocaine 2 per cent. into the anterior end of the turbinate. A second injection can often be made an inch further back along the turbinate. A fine long needle on a dental cartridge syringe is ideal for the purpose.

In several cases we have used successfully the instillation method of Curtiss (1952) using Xylotox 4 per cent. instead of cocaine. However, the technique described here proved quicker and less uncomfortable for the patient, and entirely adequate for the limited requirements of submucosal diathermy.

During the operation under local anæsthetic it is important to guard against involuntary head movements by the patient. This is best done by having him lie on a couch, with the head resting on a pillow.

Complications

Two of our cases developed adhesions between the entry point of the electrode and a marked anterior convexity of the septum. Submucous resection of the septum has now given complete relief and should have been done in the first place.

Bleeding from the puncture site has never been persistent, and is rare under local anæsthetic.

In one of our cases there was some persistent crusting, which terminated with the extrusion of a minute flake-like sequestrum of bone. Apart from this no sloughing of the turbinal bone as mentioned by previous authors has been encountered.

Clinical Material

A series of a hundred cases treated by submucosal diathermy and followed up for periods of not less than six months has been studied The cases selected for this treatment make up a very heterogeneous group No patient was rejected who suffered from longstanding nasal obstruction due to turbinal engorgement—"chronic priapism of the nose", as Richardson calls it—provided that

1 Sinus infections had been dealt with first In this series sinus radiographs were taken in twenty-four cases, and the films showed abnormalities in nine of them Pus was found at proof puncture in only two of these nine

2 Reasonable steps, such as systemic antihistamine therapy and specific desensitization (when indicated), to control allergic factors had proved unsuccessful

A number of patients in the series had associated nasal conditions such as septal deviation, polypi, and chronic hypertrophic rhinitis Some had definite skin sensitivities to inhalant allergens, others had none Of the latter many had symptoms precipitated by changes of temperature or humidity, or by emotional factors (non-specific nasal hypersensitivity) Fifty-five patients were male and forty five female Seventy-three were treated under general anæsthetic, twenty-seven under local Table I shows the ages of the patients

TABLE I

AGE GROUPS

Age in Years	Number of Cases
1–10	7
11–20	23
21–30	24
31–40	22
41–50	14
51–60	8
61–70	2

Results

A *Post-operative period* There has been little or no post-operative discomfort, apart from slight increase in nasal obstruction lasting from one to five days No special post-operative care is necessary There is a striking absence of crust formation Provided that treatment has not been excessive only a small scab is seen anteriorly where the electrode has entered the turbinate

B *Period of shrinkage* This may begin in the first three to five days as the reactionary œdema subsides There is a progressive improvement in the nasal airways which may continue for up to three or four weeks before it is complete During this time many of the worst cases of rhinitis medicamentosa have thrown away the drop bottle, and the vicious circle has at last been broken

C. *Long-term results.* The benefits are assessed in terms of relief of nasal obstruction, although some reference will be made to effects on sneezing and watery nasal discharge in Table V.

Table II shows that of ninety-eight patients complaining of obstruction, eighty-eight obtained complete or partial relief.

TABLE II

RELIEF OF NASAL OBSTRUCTION

Follow-up period in months	6–12	13–18	19–24	25–30	Over 30	Total
Number of cases completely relieved	17	11	10	2	2	42
Number of cases improved .	12	18	8	7	1	46
Number of cases unchanged ..	10					10
				Total ..	..	*98**

* The remaining two cases in the series complained only of paroxysmal sneezing and rhinorrhoea. (See Table V.)

The main interest centres on those fifty-six cases who obtained *incomplete* relief or were not improved at all. Those with incomplete relief are analysed in Table III.

TABLE III

CASES SHOWING IMPROVEMENT, BUT NOT COMPLETE RELIEF, OF NASAL OBSTRUCTION

Follow-up period in months	6–12	13–18	19–24	25–30	Over 30	Total
Cases with *improvement maintained* at the interval indicated	7	10	7	6	1	31
Cases which *relapsed* at the interval indicated .. .	5	8	1	1	0	15

Of the 15 cases which relapsed
12 were treated again with further initial success,
1 required S M R , which has given permanent relief,
1 had associated collapse of the alae nasi,
1 refused further treatment

Of the ten patients who obtained no improvement at all, no less than eight had evidence of severe nasal "allergy" and six of these had positive skin tests. In view of this we have analysed the influence of allergy upon the overall results of treatment. After excluding all cases in which obstruction was clearly contributed to by septal deviation needing resection eighty-seven cases remain. These are classified in Table IV.

TABLE IV

INFLUENCE OF NASAL ALLERGY ON RESULTS

	Completely Relieved	Improved		No Change
		Maintained	Relapsed	
Cases with clinical impression of allergy and skin tests positive	3	6	3	6
Cases with clinical impression of allergy, but skin tests negative	9	8	3	1
Cases with no evidence of allergy	26	14	6	2

This shows that cases having positive skin tests have the highest failure rate The best results were obtained in cases with no evidence of "allergy' as generally understood in the clinical sense

All of the foregoing data relate to nasal obstruction Relief of watery discharge and sneezing occurred as shown in Table V

TABLE V

RELIEF OF WATERY NASAL DISCHARGE AND SNEEZING

Complaint	Cases completely relieved	Cases improved	Cases unchanged
Watery nasal discharge 57* cases	9	16	32*
Sneezing 48* cases	8	10	30*

* These figures include the two cases who had these symptoms prior to operation but did not complain of obstruction

These findings confirm that relief of sneezing and watery discharge is unpredictable It is difficult to understand why any improvement in these symptoms should result from submucosal diathermy We agree with Shahinian who was "reluctant to advise submucosal diathermy unless the patient feels that the prospect of relief of nasal obstruction would in itself be sufficient reason for undergoing the procedure"

Associated Conditions Requiring Additional Treatment

Nasal polypi were removed in six cases at the time of performing submucosal diathermy with satisfactory results

Deviated Septum requiring submucous resection was present in thirteen cases In most of these submucosal diathermy was used to reduce a compensatory turbinal hypertrophy to conform with the corrected position of the septum.

Chronic (suppurative) sinusitis was present in two cases, one being associated with bronchiectasis.

Tonsillectomy was performed at the time of submucosal diathermy in two cases, and adenoidectomy in one of these.

Chronic hypertrophic rhinitis was present in nine cases, with morelliform enlargement of the inferior turbinate localized to the posterior end or lower border. It was considered in each of these cases that submucosal diathermy alone would be inadequate to reduce the localized area of hypertrophy, although the co-existing diffuse turbinal engorgement was suitable for diathermy. The latter was therefore supplemented by surgical reduction in these cases. Five of them obtained complete relief of obstruction, and four showed improvement.

Summary

A new instrument and a technique are described for submucosal diathermy of the inferior turbinate, and the literature is reviewed.

The method, which is equally suited by local or general anæsthesia, is considered superior to linear galvano-cautery of the inferior turbinate, in that it is less injurious to the epithelial lining of the nose.

The results of treatment in a series of a hundred cases are given. Of ninety-eight with chronic turbinal engorgement who complained of nasal obstruction forty-two obtained complete relief, and forty-six were improved. Better results were obtained in the "non-allergic" than in the "allergic" group.

The method can be combined with the treatment of associated conditions such as polypi, septal deviations, or chronic hypertrophic rhinitis.

Acknowledgments

We are grateful to Mr. Ian G. Robin for allowing us to include a number of his cases in this series, and to Miss Diane Hendley for invaluable help in organizing the follow-up arrangements. We are greatly indebted to Mr. W. Whittlesea for his energetic help and constructive criticisms in the design of the instrument. The Photographic Department of St. Mary's Hospital kindly prepared the photograph.

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HÆMOLYMPHANGIOMA OF THE NECK

By E. A. MALKIN (London)

THIS case is described, not so much because the pathology is relatively uncommon in the neck, but because of the surgical difficulties encountered, and the cardiac incident which provided the final episode. It is worthy of mention, that in all the procedures which will be recounted, the morale of the patient and his astonishing insight never failed him for a moment; he was an extraordinary man in more ways than one.

His story begins in 1920, when he was admitted under Sir Launcelot Barrington Ward at the Hospital for Sick Children, Great Ormond Street, with a nævus of the right side of his face and neck. This had been first noticed at the age of six months, and had been cauterized twice in the intervening period. The lesion was again cauterized on more than one occasion, and by the time he was four years old had spread as a papillomatous nævus from the right cheek down to the shoulder on that side, and had involved the right eyelid and an area around the mouth and nose.

In 1937, as a student aged 19, he was admitted to The General Infirmary at Leeds when he complained of dysphagia. There was a large swelling of the neck which moved on swallowing, and nævi of the neck, mouth, and right eyelid. A subtotal thyroidectomy was performed, and the specimen found to contain two large swellings, one very cellular, the other adenomatous.

In December, 1948, he was seen by Dr. C. J. Gavey at The Westminster Hospital when he complained of cough and epigastric discomfort, but no significant pathology was found in the digestive tract. He was referred to Sir Stanford Cade regarding the condition in his neck, who commented: "The patient has an enormous congenital hæmangioma of the neck, involving mainly the right side of the thyroid gland, but extending from the gland right up to the skin, and also to the mucosa of the pharynx. There are also large masses of papillary lymphangiomatous tissue on the right side of the buccal mucosa of the lower lip and on the back of the tongue, and that there is also a capillary hæmangioma in the inner canthus of the right eye. The right side of the face is hypertrophied due to the gross vascularity present since birth."

It was not considered that any treatment, either by surgery or radiation was indicated, since the condition had remained stationary for a decade, and caused him little disability. Indeed at that time he was an active tennis player, and subsequently in 1952 he was passed fit for duties in America, although he did not in fact cross the Atlantic

He was next seen in February, 1955, when he complained of slight hæmoptyses associated with exacerbations of his chronic cough His breathing was noticed to be noisy, and there was slight dyspnœa on exertion Chest skiagrams did not reveal any mediastinal extension of the tumour, but there was some dilatation of the superior vena cava The heart size was normal The trachea was displaced to the left, and compressed from side to side

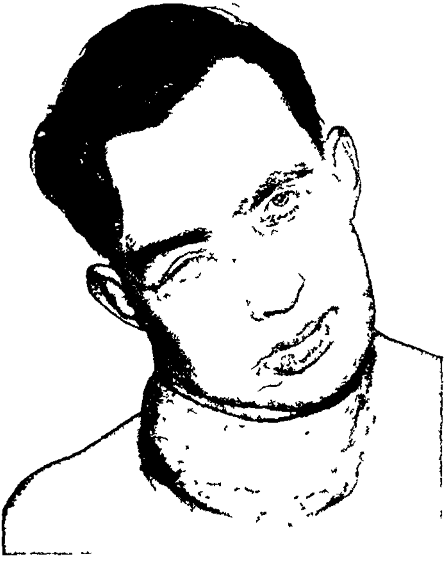

Fig 1

He also complained at this time of pain in the right eye, which was found to be slightly proptosed, and to have some limitation of depression and abduction There was considerable œdema of the lids, and the tumour of the upper lid at the inner canthus was larger than hitherto This mass was excised and found to be a lymphangioma

On January 9th, 1957, he was admitted as an emergency with stridor at rest.

His cough had become very troublesome during the previous month, and this with dyspnœa and difficulty in expectoration has led to a gross loss of sleep and some mental confusion at night. The swelling then involved the whole of the right side of the neck, and extended from the lower jaw to the clavicle, and also had spread to a lesser degree to the left side as well (Fig. 1). The mass was "doughy" in consistency, and moved on swallowing. There was a bruit, and transmitted pulsation on the right side. It was impossible to obtain a view of his vocal cords by indirect laryngoscopy.

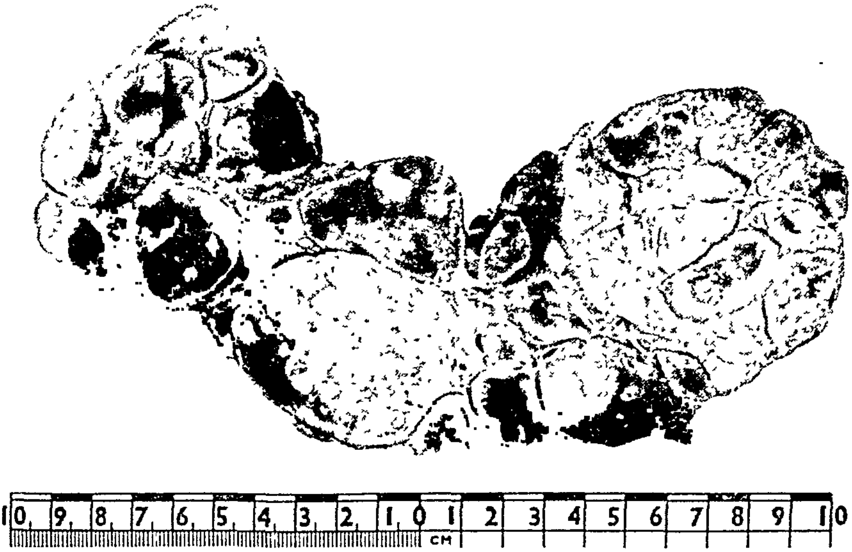

FIG. 2
Operative specimen: nodular goitre.

At this time, skiagrams showed a mediastinal shadow on the right side, presumably due to an extension of the lymphangioma, while the œsophagus was displaced forward at the level of the thyroid. The heart was also enlarged, and the right diaphragm paralysed.

By January 17th his dyspnœa, chiefly inspiratory, was causing him considerable distress, and some attempt to relieve the obstruction seemed imperative.

An operation was therefore performed (Sir Stanford Cade and Mr. Miles Foxen) when an incision was made over the left aspect of the tumour and extending just across the midline to the right side; there was very considerable hæmorrhage from large dilated veins. It was apparent that a considerable proportion of the mass was due to an enlarged thyroid gland, and this was excised with some difficulty, together with some extra hæmolymphangiomatous tissue. A high tracheostomy was made, and a rubber tracheostomy tube inserted

On recovery from the anæsthetic in the ward, the patient had a bout of coughing which resulted in an alarming hæmorrhage from the incision. He was taken back to the theatre where the bleeding points were ligatured (E.H.M F.) and a cuffed tube inserted through the tracheostomy. Nine pints of blood were given during these procedures.

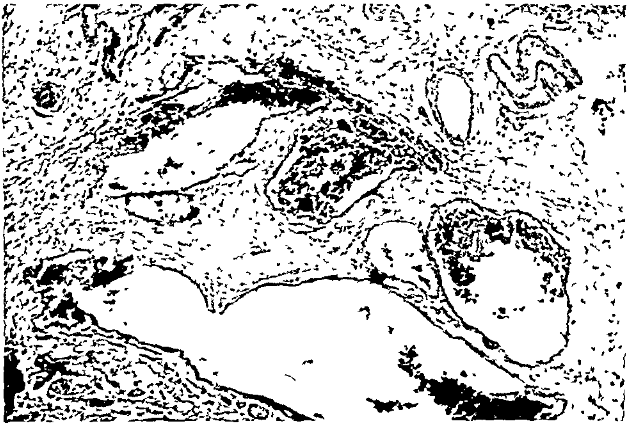

Fig 3

Post-mortem specimen × 24 showing cavernous space in the neck tumour

He had not fully recovered consciousness by the next morning, although he had been kept in an oxygen tent. A remarkably quick improvement resulted, within minutes, by changing the cuffed tube for a large Edinburgh pattern one. Presumably, the cuffed tube had been inserted into the right main bronchus, served, if that be the term, by the paralysed right diaphragm.

The operative specimen (Fig. 2) consisted of a mass weighing 398 grams. The external appearance was that of a nodular goitre, and the cut surface showed pink fleshy areas and multiple cystic areas. Histologically, the appearances of the main mass were that of a nodular goitre. Fig. 3.

His progress during the ensuing month was relatively uneventful except for slight bleeding from the tracheostomy. This necessitated rather frequent changes of the largest sized Durham's tube, which was found to be the most comfortable type of tube. This bleeding was felt to preclude the removal of the tube, although his natural airway was reasonable in spite of the bilateral cord palsy which had been confirmed by direct laryngoscopy early in February.

He was discharged on March 5th, but had to be readmitted for eight days on the 9th, on account of further bleeding around the tube.

He was readmitted on April 30th for reassessment. The bleeding from the tracheostome track, itself 3 in. deep, was still proving troublesome, and on one occasion amounted to 10 oz. The measures adopted to control this hæmorrhage, namely morphia and local cauterization, were proving less and less effective as time went on. Fortunately most of the bleeding was towards the surface, and not into the trachea. The tube was therefore removed, although it was realized that if severe dyspnœa resulted, then urgent and difficult surgery would be needed. Furthermore, if the bleeding did not cease as a result of removing the irritation due to the tube, then the blood might well travel into the trachea, and prove impossible to deal with. After ten days of some anxiety, with intermittent and slight bleeding, the fistula was closed, and the patient sent home.

He was again admitted on May 18th with trembling of the hands and feet, and considerable inspiratory obstruction. There was no cyanosis and the patient was alert and co-operative. The picture seemed to be that of relative anoxia and parathyroid deficiency. By the 21st of the month, stridor was present throughout the day and night, and sleep was disturbed and fitful. Skiagrams revealed that the heart had increased in size yet further, and that the right diaphragm was even higher than before.

He was seen by Dr. C. J. Gavey, and Sir Stanford Cade on several occasions, and it was decided to explore the neck with a view to reconstituting the tracheostomy in such a manner that the skin edges would be brought to the margins of the actual tracheostome; it was also hoped that a significant part of the tumour which remained might be removed at the same time.

This was planned for May 25th, but on the 23rd there was a sudden deterioration with increasing dyspnœa and mental confusion, which rapidly extended to stupor. A nasal endotracheal tube was passed while the patient was in bed (Dr. G. Organe) but this did not effect the level of his stupor.

Later that day (Sir Stanford Cade; Mr. Miles Foxen) a transverse incision excising the original scar resulted in so much hæmorrhage, that it was immediately apparent that there was no hope of achieving more than a tracheostomy. Attempts to reach the trachea just above the sternal notch, and at the level of the cricoid, were unsuccessful due to profuse bleeding A third attempt at the level of the third ring was successful, and the skin edges were sewed to the edges of the tracheal opening

At this juncture it was noticed that his pulse was impalpable, and that no heart beat could be heard It is difficult to be certain how long elapsed between the actual commencement of the cardiac arrest, and the thoracic incision (E H M F) but it may well have been two or three minutes The heart was atonic, and not fibrillating, 1 c ml of 1/1,000 adrenalin was injected into the left ventricle after manual massage of the heart had been commenced, and the heart beat returned after five minutes

Spontaneous breathing had also ceased, so he was transferred to the ward and respired by a positive pressure machine

The next day he was breathing spontaneously, his blood pressure was around 110/70 but the radial pulse was sometimes impalpable There was then a period of cerebral irritation with strong convulsive seizures An E C G did not show any significant abnormality After eight hours it was decided to try and control the convulsions by intramuscular paraldehyde, local peripheral cooling and by the central effects of chlorpromazine, 25 mg six-hourly The convulsions certainly decreased in intensity, but by the evening of May 25th his blood pressure had fallen to a very low level, and he died at 10 p m

Post mortem revealed early necrobiotic changes in the cerebral cortex and brain stem, indicative of an arrested circulation There was gross hypertrophy of the left ventricle of the heart the heart weighed 22 oz The trachea was deviated to the left by a diffuse hæmolymphangiomatous mass involving the whole neck and there was some extension of the tumour into the upper mediastinum, although rather less than had been anticipated

The lungs were remarkably free from petechiae, but the lower bronchial tree was filled with mucopus considered to be largely terminal There were no tumour masses in the brain, liver, or gut Fig 3 shows a post-mortem section of the cavernous hæmolymphangiomatous spaces found in the tissues of the neck

Comment

Tumours of ' vasoformative" origin present aspects of some interest, first the clinical classification varies from author to author secondly the exact pathogenesis is a matter of dispute, and thirdly the multiplicity of treatments advocated is an indication of the difficulty in dealing with them

It was Wagner (1877) who first suggested dividing lymphangiomas into three large groups simple or capillary, cavernous, and the cystic hygromata. Even today, this division would appear to satisfy many writers so far as the simple lymphangiomata are concerned. The hæmangiomas are similarly described by some workers, but the clinical types are so varied that it is probably justified to subdivide the group.

The dispute over the pathogenesis of these tumours centres around whether they should be regarded as true new growths, or whether they would be better placed with the Hamartomata: congenital tumour-like malformations in which tissues of a part are present in improper proportions or distribution (Albrecht, 1904).

The clinical manifestations of these tumours vary from small localized superficial tumours of the skin or mucosa to diffuse masses involving much of the face and neck, or even whole extremities. They may indeed occur in a multiple form such as Lindau, von Hippel's disease where hæmangioma of the retina is associated with cerebellar angiomata and cysts of the pancreas, spleen and adrenal. Another multiple syndrome is Sturge Weber disease in which an angioma follows the distribution of the trigeminal nerve, and further lesions are found in the brain and orbit.

In an interesting report on 1,056 cases, Watson and McCarthy showed that of a total of 1,363 lesions, cavernous angiomas accounted for 952, capillary angiomas 291, pure lymphangiomas 41, cystic hygromas 14, while all the other clinical subdivisions accounted for the remaining 65. There was evidence of their presence at birth in 73 per cent. of the cases, and a further 12 per cent. had appeared by the end of the first year; furthermore, they constituted the largest single group of childhood neoplasms. The overall distribution was: head 52 per cent., neck 4 per cent., trunk 23 per cent., extremities 19 per cent., and genitalia 2 per cent. They are more common in the female, which sex accounted for 65 per cent. in this series.

Apart from those cases where the tumour is clearly a lymphangioma or an hæmangioma, many cases are found where elements of both types are present. Apparently, hæmangiomatous elements are more likely to be found superimposed upon a lymphangioma, than the other way round (Havens and Lockhart, 1953).

In the vast majority of cases angiomas are benign in nature, although they may be responsible for distressing disfiguration. Many enlarge *pari passu* with the growth of the patient, and cease to increase in size when adult life is reached. Very occasionally however, an angioma may suddenly take on aggressive tendencies and rapidly become much larger. Conversely, it is not unknown for small angiomas to regress: Ribbert showed by injection technique that most small angiomas have only one afferent and one efferent vessel, and it is generally accepted that an accidental thrombotic occlusion of the afferent vessel accounts for these cases of spontaneous regression. A few cases have been described where multiple successive growths have occurred, some with a fatal ending. It is a matter of dispute whether these are to be regarded as examples of metastatic or of multicentric growth

The following classification embraces most of the conditions met with

Hæmangiomata

Capillary where the basic characteristic component is a thin walled endothelial tube In some, areas of solid endothelial masses are also included presumably masses of angioblastic cells which have not canalized

They include the strawberry and salmon patches, port wine stain and the spider angiomata

Cavernous where the blood spaces are large, irregular, and intercommunicating, *and with a resemblance to erectile tissue The lining cells are* flattened, and there is little endothelial matrix, while the supporting tissue consists of thin fibrous intervascular septa, and some smooth muscle They are often associated with an overlying capillary lesion

They are found not only in the skin, but in the liver, spleen, kidney, breast and tongue

Hypertrophic where the endothelial matrix predominates, and the patent capillaries few in number They tend to be locally invasive

Racemose where a soft pulsating compressible tumour, capable of eroding bone is found This type probably includes tumours of more than one origin The circoid aneurysm of the temporal artery is one type, while a rare plexiform angioma of the head, neck and forearm composed of arteries, veins and capillaries is another

Hereditary hæmorrhagic telangiectasia This has recently been described by Harrison, 1957, and is typified by multiple telangiectasia found in the skin and in the mucous membranes of the nose, pharynx, lips and elsewhere An erudite account of telangiectatic conditions has been given by Parkes Weber, 1947

Diffuse Systemic This is usually a mixed hæmolymphangiomatous lesion involving a limb, and would appear to have grown with the developing limb bud

Metastasizing A few well authenticated cases have been reported, where multiple and apparently successive growths have occurred, involving the skin and viscera Some of these have terminated fatally Borrman, 1906, described the first case in the literature, while more recently Robinson and Castleman, 1936, have described another case and reviewed the subject

Lymphangiomata

These are very much less common than angiomas, and morphologically may be described as capillary, cavernous, or cystic These latter cases have been very well and fully described by Goetsch, 1938

The discussion as to the underlying nature of these tumours occupies much of the extensive literature on these conditions It is necessary, of course, always to exclude vascular reparative tissue which may mimic a true angioma This has recently been emphasized in the case of some laryngeal angiomata (Stewart, 1957).

It is now generally accepted that the vascular system arises *in situ* throughout the mesenchyme of the body by a coalescence of plexiform clefts from primitive "blood island" angioblasts. This theory is in contrast to the original idea of His which invoked a continuous spread of vascular tissue from the extra-embryonic yolk sac.

The persistence of isolated areas of vascular tissue which fail to join up with the rest of the system would be a possible cause of congenital malformation. On the other hand, it appears that some authorities believe that capillary endothelium may form sprouts which subsequently canalize to bring about a true neoplastic formation.

Harvey, Dawson and Innes, 1940, in a superbly illustrated article, include the tumours under discussion in the endothelioma, more particularly as hæmic or lymphatic vasoformative endotheliocytoma . . . the suffix referring to an incapacity for proliferation on the part of the endothelial lining. They stress the fact, that it is often not possible to lay down where an angiectatic malformation ends and an angiomatous tumour begins. However, as Evans, 1956, has pointed out, cell differentiation does not necessarily imply an inability to undergo neoplasia. Gruenwald, 1943, states that "no morphological terms and characteristics can serve to indicate the developmental potencies of cells . . . and that dysontogenetic tumour formation does not necessarily imply an embryonic anomaly".

Willis, 1948, is a strong protagonist of the belief that the majority of cutaneous angiomata are developmental anomalies, citing that in general they are present at birth, that they do not involve a greater and greater territory after growth ceases, and that they usually remain unchanged unless accidents such as trauma, thrombosis, or infection supervene. In brief, he believes that they have no power of disproportionate growth. He also points out that the diverse potentiality of proliferating mesenchyme makes the sharp differentiation of angiomas from other mesenchymal growths to some extent arbitrary. He accepts, however, that there are a few cases of true malignant neoplastic angiomas.

A very full and oft quoted account of the cystic lymphangiomata is that of Goetsch, 1938. He quotes Dowd, 1913, who accepted the view that hygromata arise from sequestrations of lymphatic tissue derived from the primitive lymph sacs, and he believes that these sequestrations possess independent powers of original growth by the budding off of endothelial sprouts which subsequently become canalized, and that consequently these tumours are neoplastic.

A great variety of treatments have been advocated for these conditions, indicative of the difficulty in dealing with them. The following scheme outlines the scope of treatment:

(*a*) Surgical removal;

(*b*) Ligature of the afferent vessel;

(*c*) Obliteration of component vessels by:

1. Refrigeration; CO_2 snow.
2. Sclerosants:
   Ethamolin. Sodium morrhuate, boiling water, concentrated saline.
3. Radiotherapy.
   X-rays, radium, radon.
4. Deep electrocoagulation.

The cutaneous lesions give rise to a cosmetic problem, and their treatment has been fully outlined in a recent paper, Matthews, 1951.

The diffuse cavernous conditions present a great problem, more especially when they are too large for sclerosant therapy, and where the possibility of total surgical removal is remote. In earlier days there was a heavy operative mortality: Dowd, quoting Farr's statistics, showed that of 91 cases of cystic hygromata, 39 died while under treatment, 23 following excision or puncture. More recently, however, Goetsch had only one death in twelve cases operated upon, and that was in an infant aged six months where the tumour was larger than the patient's head

My thanks are due to Sir Stanford Cade for permission to describe his case, and to Dr. P. Hansel and Dr. D. McKenzie for the illustrations. I would also acknowledge the early information obtained from The Hospital for Sick Children, Great Ormond Street, and from The General Infirmary at Leeds

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ACUTE OSTEOMYELITIS OF THE MAXILLA IN INFANTS

By A. E. W. GREGSON (Leeds)

In 1939 Asherson had seen only four examples of this condition in the preceding ten years. The occurrence of six cases in the Leeds area between 1953 and 1956, at a time when a wide range of antibiotic agents was available, has provided an unusual opportunity of assessing the value of these drugs and of considering how their use has influenced the pattern of therapy.

Since this series was not anticipated, no policy had been laid down with regard to treatment. Each patient was treated in the light of the evidence available at the time. A review at this juncture is therefore not inapt, for, in the words of Oscar Wilde, "experience is the name we all give to the recognition of our mistakes".

The symptoms and signs of the disorder are not discussed; little could be added to previous accounts. The purpose has been to rationalize and elucidate the problems presented in the treatment of this infection, with particular reference to its features as a staphylococcal invasion. The facility with which this organism develops resistance to antibiotics is now well known, and is almost a subject with its own bibliography.

Pathology

No study of therapy is adequate without a preliminary consideration of the pathology of the condition. Details of the morbid histology, especially in the early stages of infection, are scanty, and, in the absence of any mortality in the present series, it has not been possible to add to the present small store of knowledge. Nevertheless, so many theories have been advanced in the past that further conjecture is at least in line with previous practice.

Zarfl (1920) advanced the theory that a stomatitis was the cause of the condition. Bass (1928), Asherson (1939) and McCash and Rowe (1953), have all supported the view that the lesion was primarily an infection of the tooth-bud, the last-named mentioning specifically the first molar tooth. Lillie (1946) advocated the tear sac as site of primary invasion. Poncher and Blayney (1934) and Howarth (1947) implicated the maxillary antrum, although Asherson dismissed this possibility.

In 1932, Wilensky expressed the view that "osteomyelitis of this (maxillary) type occurring in nurslings and sucklings is in no way different from the hæmatogenous form of osteomyelitis occurring in other parts and bones of the body and that the occurrence of the lesion in such young subjects is associated with the physical conditions of childbirth and the environmental conditions immediately following" Asherson also believes that the infecting organism reaches the maxilla by the blood stream but Macbeth (1952) disputes the view However, the "physical conditions of childbirth and the environmental conditions immediately following" provide no reason for the occurrence of infection at the age of fifteen months, as in Case VI of this series, or even later as in one of Macbeth's cases

The striking feature of this disease is, of course, its manifestation at a particular age, between birth and three years old, and it has seemed logical, therefore, to consider the way in which the maxilla differs physiologically from other bones at the same age and from itself in later years The characteristic which immediately becomes apparent is the remarkable degree of dentigenic activity At birth, calcification is apparent in all the tooth buds of the deciduous eruption, together with the central incisor, canine, and first molar of the permanent dentition (Kronfeld, 1939) The same process is going on in the mandible, but there it is not condensed into so small a volume By comparison, growth activity elsewhere in the skeleton is relatively small

This process of calcification is associated with two combined conditions in the local circulation, hyperæmia and relative stasis The circulatory state of the maxilla at this age is, therefore, comparable with that existing at a later age in the annulus vasculosus at the metaphysis of a long bone, and in both situations the conditions are ideal for the lodgement of septic emboli Since calcification is proceeding more rapidly in the maxilla than anywhere else during the first year of life, it is only to be expected that this will be the most likely site in bone where these emboli will settle

The source of the emboli is immaterial They may be from pyogenic lesions elsewhere, and co-existing lesions have been described quite frequently (Bass, 1928, Lacy and Engel, 1939, Cohen, 1949) The staphylococcus may be harboured in the child's nasopharynx or elsewhere Martyn found the organism present during the first week of life in 76 per cent of children born in a Maternity Unit (Martyn, 1949) The important feature is that, if staphylococci gain access to the blood stream, the maxilla is, at this age, the most likely bone to show abscess formation

With increasing age, the dentigenic activity slows down, and by the end of the first year has already abated considerably The bones of the limbs are beginning to take over as the sites of maximal growth activity and, therefore, of most rapid calcification Osteomyelitis is then much less likely in the maxilla and becomes more frequent in the limb bones

Case VI of this series is put forward as an example of the stage of transition. He developed infective foci almost simultaneously in the maxilla and the calcaneum.

The picture is primarily one of septicæmia, and the fact that local manifestations occur particularly in the maxilla is of no more significance than local manifestations of infection elsewhere would be. If, however, the maxillary invasion is not adequately dealt with, the septic thrombosis will give rise to devitalization of bone and the subsequent formation of sequestra, and sequestra at this site may present peculiar problems of treatment.

Case Histories

1. D.A.H., male, aged five weeks.

First seen 6.2.53. A swelling of the right side of the face had been noticed by the parents for two days. This was seen to be lateral to the nose and below the eye, red and tender, but not fluctuant.

Pus was seen in the right anterior naris. The temperature was 102° F. on admission.

He was given 500,000 u. soluble penicillin immediately followed by 150,000 u. distaquaine twice daily.

After 48 hours there was slight reduction of the pyrexia, but no improvement in the swelling, which had become fluctuant. The swelling was incised under general anæsthesia and pus was released containing *Staph aureus*, sensitive only to streptomycin and aureomycin. When the sensitivity report was available (5th day), the child was given 50 mg. aureomycin every six hours by mouth. On the seventh day an incision in the canine fossa released more pus; twenty-four hours later the child was apyrexial, and the swelling of the cheek had almost disappeared. Aureomycin therapy was continued for a total of fourteen days and the child was kept under observation for a further seven days. He was last seen fifteen months after the onset of trouble, and up to that time had suffered no more trouble.

Commentary. Clinically it was apparent that the staphylococcus was resistant to penicillin before a report of bacteriological sensitivity was available. Had a specimen of the nasal pus been examined initially, the most suitable antibiotic could have been given at least three days earlier. In this particular case no ill-effects resulted from the delay.

2. M.F.P., male, aged six weeks.

First seen 7.10.54. Watering of the left eye and obstruction of the left side of nose six days before. Swelling of the left orbit, cheek, and side of nose twelve hours later. Intramuscular penicillin therapy had been given by the family practitioner. After a slight initial improvement proptosis of the left eye and a purulent nasal discharge developed.

On admission the temperature was 99° F., there was proptosis of the left eye, but no chemosis; the left cheek was swollen, and pus was seen in the left anterior naris. No bacterial growth was obtained from a culture of the nasal discharge. The child was treated with penicillin 500,000 u. six-hourly and streptomycin 0·25 gm b.d. both by intramuscular injection. After five days the child was apyrexial, and the swelling of the cheek was settling. Therapy was stopped three days later, and the child returned home after a further three days.

Two weeks later the condition flared up again, but responded to daily injections of penicillin 500,000 u. During this episode the child was treated at home by the family practitioner.

Some twelve months later the left side of face and orbit began to swell from time to time. These attacks settled without treatment. Radiographs were taken of the left maxilla, and it is possible that one of them shows a sequestrum (see Fig. 1).

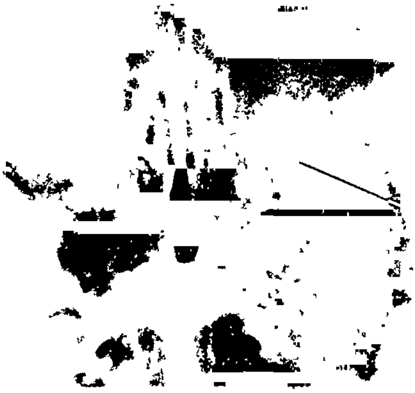

Fig 1

Case 2 Radiograph taken twelve months after the onset of infection shortly before a sequestrum was extruded

Eighteen months after the initial illness a small sequestrum was extruded through the skin below the left inferior tarsus.

The child has remained well since then.

Commentary. It seems likely that penicillin had at least partly controlled the infection before the child was seen in hospital. When resolution was incomplete no organism could be detected by culture, so that any alteration in therapy had to be made without adequate bacteriological guidance Since penicillin effectively cleared the exacerbation two months later the organism must have remained sensitive to that drug.

Sequestrum formation occurred, but gave little trouble, since the devitalized fragment was extruded spontaneously. Any attempt to remove it by surgical exploration could have caused scar formation with a much more unsatisfactory cosmetic result.

3. A.G.S., male, aged three weeks.

First seen 11.2.55. Two weeks purulent right nasal discharge. An erupted tooth was present at birth in the right upper alveolus, and came out shortly afterwards. The parents had been advised to massage the exposed tooth socket with some unidentifiable medication.

Shortly before admission, swelling and inflammation of the right cheek developed. On examination pus was seen in the right anterior naris. Pus was draining from the tooth socket in the right upper alveolus. There was a brawny inflammatory swelling of the right cheek. The child was given penicillin 500,000 u. six-hourly by intramuscular injection.

After 24 hours the child did not appear acutely ill, but a low-grade (99° F.) pyrexia persisted. Unfortunately, bacteriological examination of the nasal and dental discharges was delayed, and a report was not available until late in the second week of treatment. *Staph. aureus* was present in cultures from both sites, resistant to penicillin, and most sensitive to aureomycin. Penicillin therapy was stopped, and the child was given aureomycin 40 mm. six-hourly orally. After seven days of this treatment the discharge from the maxillary alveolus ceased, and two days later the nasal discharge also ceased.

The child was reviewed two weeks later, and was quite well. Unfortunately the parents failed to bring him for further review.

Commentary. Again bacteriological investigation was not done early enough. As soon as it was carried out, and the correct antibiotic was applied rapid resolution occurred.

4. A.G., female, aged three weeks.

First seen on 5.8.55. The parents had noticed watering of the right eye and swelling of the right cheek that day. On examination, there was swelling of the right orbit with proptosis of the eye. The right cheek was swollen, and pus was present in the right anterior naris. The child was given penicillin 250,000 u. six-hourly by intramuscular injection.

Although the temperature settled slowly, the swelling of the right orbit and cheek persisted, and, after four days, papillœdema was present in the right fundus. An incision down to bone was made below the right eye to relieve the orbital tension. Although frank pus was not released by this incision, the blood-stained discharge contained *Stapah. ureus* which was resistant to penicillin, but sensitive to streptomycin.

When this report was available the child was given streptomycin 0·25 gm. six-hourly by intramuscular injection. The papillœdema had resolved twenty-four hours after incision and the swelling of the orbit and cheek was settling. Seven days later the swelling had gone completely, there was no proptosis, and the nasal discharge had ceased.

The child was last seen on 1.7.57, some two years after the initial symptoms, and up to that time had remained well.

Commentary. Bacteriological investigation could have been carried out earlier upon the nasal discharge, with consequent earlier application of the most suitable antibiotic, and possibly the avoidance of papillœdema. When papillœdema had developed, an incision to reduce the orbital tension which had caused it gave rapid relief.

Once the most suitable antibiotic was given rapid resolution occurred.

5. J.C., male, aged one month.

First seen on 17.1.56. He had been given penicillin for a swelling of the right orbit for three days, and terramycin for two days. There was swelling of the right periorbital tissues and cheek, and pus was escaping from a fistula in the right upper alveolus. The pus contained *Staph. aureus*, sensitive *in vitro* to penicillin, streptomycin, aureomycin, chloromycetin, terramycin and erythromycin.

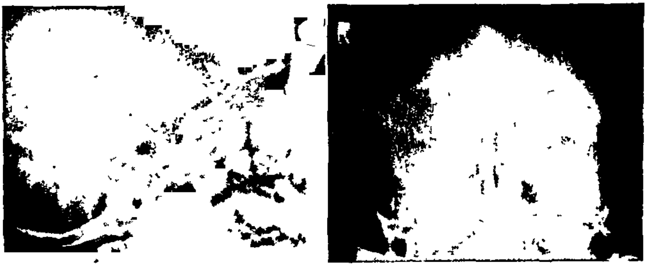

FIG 2

Case 5 Radiographs taken nine months after the onset of infection Although the antrum on the affected side is opaque, there is no evidence of a sequestrum

The terramycin therapy was continued, and after twenty-four hours pus was drained by an incision below the lower eyelid. Progress thereafter was uneventful, and antibiotic therapy was stopped after seven days, the child being allowed to go home.

He was readmitted two weeks later, with pyrexia and a recurrence of the swelling of the right cheek. He was given penicillin by intramuscular injection (200,000 units distaquaine daily). After one week the lesion seemed to have settled, and therapy was stopped. The swelling reappeared, however, one week later still, and it was noticed that pressure over it caused a flow of pus from the anterior naris He was again given penicillin in the same dosage. The swelling settled after fourteen days treatment, and the child was allowed to go home.

Some six months later, a reappearance of the swelling was treated by penicillin therapy in the same manner, and resolution occurred after nine days.

Nine months after the child was first seen, the swelling recurred yet again, being most dependent below the outer canthus of the eye. He was given penicillin again in the same dosage and the swelling was incised below the outer canthus of the eye. Pus, containing *Staph. aureus*, was released, but antibiotic sensitivity was not assessed. The swelling settled in two weeks, and there has been no further trouble. A radiograph of the right maxilla nine months after the initial infection showed no evidence of bone infection (Fig. 2), although the antrum on the affected side was opaque.

Commentary. The initial therapy was probably terminated too soon, allowing the staphylococcus to regain its hold.

The later recrudescences may have been associated with either a residual abscess or the formation of a sequestrum. No radiological evidence of a sequestrum was found.

The continued efficiency of penicillin therapy in the later recrudescences is noteworthy. Theoretically penicillin resistance was to be expected.

6. D.C., male, aged 15 months (Gregson and Yeoman, 1957). Previously published as a Case Report in *J. Laryng.* (Oct., 1957) by the writer, in collaboration with P. M. Yeoman.

First seen 16.6.56. Swelling of the right orbit, cheek, and upper lip had been present for two days, and the parents thought that there had been a "septic spot" on the upper lip four days earlier. Temperature 100·5° F. Apart from the swelling described, the child was obviously ill, and the mouth was dirty, although no dental sepsis was seen. He was given 200,000 units of penicillin intramuscularly every six hours.

In the next twenty-four hours his condition deteriorated and he became severely toxic, with a temperature of 104° F. There was a proptosis and ophthalmoplegia of the right eye, a fluctuant inflammatory swelling of the right cheek, and three small areas of cellulitis had appeared, one on each side of the chest, and one over the lower part of the left leg. A specimen of blood, taken for culture, showed a heavy growth of *Staph. aureus*, resistant to penicillin but sensitive to erythromycin. Penicillin therapy had been stopped when the deterioration became apparent, and erythromycin 100 mg. orally every six hours had been substituted.

So that the ophthalmoplegia might be relieved the swelling was incised below the lower eyelid and a small quantity of pus was released, a rubber drain then being placed in the wound. This pus also contained *Staph. aureus*, sensitive to erythromycin.

Twenty-four hours later there was a general improvement, but the child resented any examination of the left ankle, which was hot and swollen. The leg was immobilized.

At this stage there was radiographic evidence of an opacity of the right maxillary antrum, without definite bony involvement, but films of the lungs, mediastinum, left tibia, fibula and tarsus, were normal.

After ten days the pyrexia had abated and the maxillary and orbital swelling had gone. The swellings of the right chest and left leg had settled; that of the left chest wall was fluctuant, and it was aspirated yielding a small amount of pus, containing *Staph. aureus* sensitive to erythromycin.

After twenty-five days, during which time erythromycin therapy had been continued, all the lesions had resolved except that of the left ankle. A small collection of pus had occurred at the site of the cheek incision, but it cleared up rapidly after re-opening. A small abscess of the heel was drained, and the pus from both these lesions contained *Staph. aureus*, sensitive to erythromycin.

Radiography of the left ankle, after two months, showed an area of bone destruction in the anterior part of the calcaneum, and when a sinus developed on the outer side of the heel a week later, the leg was again immobilized. An intermittent discharge occurred from the infra-orbital scar from time to time, and the surrounding inflammatory reaction caused an ectropion The discharge ceased four months after the original onset of infection, and the ectropion gradually improved thereafter.

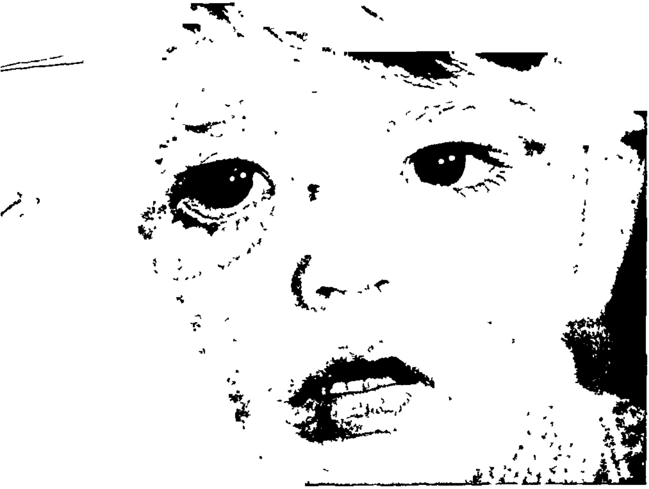

FIG 3

Case 6 One year after the onset of infection, showing the minimal ectropion on the right side The dark shadows lateral to the left eye were due to an abrasion from a fall, two days before the plate was made

A further episode of metastatic infection in the left arm developed six months after the original onset of infection, but subsided after further erythromycin therapy and immobilization of the arm in plaster

Two months later still a small sequestrum was extruded from the right maxilla into the bucco-gingival sulcus above the lateral incision Radiography of the maxilla had been done frequently before this, but no sequestrum was seen in any film. After the appearance of the sequestrum, all the lesions have resolved A slight ectropion persists, and the possibility of its correction by V-Y-plasty is being considered (Fig 3).

Commentary. This child was by far the most gravely ill of the whole series, and, but for the successful "blind" use of erythromycin, would probably have died from the initial septicæmia. The continuing sensitivity of the staphylococcus to erythromycin, in circumstances where resistance should in theory have developed, is noteworthy. This is the only example in the series where any deformity (ectropion) has resulted from the infection. Even so, the correction of the ectropion, if it is necessary, should present little difficulty.

The age of onset is greater than usual, although two years less than that of one case reported by Macbeth (1952).

Treatment

The treatment of this condition falls into two parts. First, the septicæmia must be eliminated. Later, when the generalized toxicity has been controlled, any problems presented by sequestrum formation must be solved.

The Stage of Septicæmia. In this series, antibiotics, assisted by the local drainage of collections of pus, eliminated the septicæmia effectively. At times there was some delay in finding the most suitable agent, and one is forced to the conclusion that steps must be taken as soon as the patient is seen to identify the strain of staphylococcus and assess its sensitivity to antibiotics.

Several small pieces of evidence may be useful. The presence of staphylococcal infections elsewhere either in the child, its parents, or nurse, and their response to antibiotic therapy, may indicate that some agent is particularly suitable, or that it is likely to be ineffective. At the usual age of presentation, four to six weeks, the place of birth may have significance. The child born at home is more likely to harbour staphylococci which are sensitive to penicillin than is the child born in a maternity unit (Edmunds, Elias-Jones, Forfar, and Balf, 1955) (Table I).

TABLE I

From Edmunds et al. "Pathogenic Staphylococci in the Environment of the Newborn" (1955), *B. M.J.*, i, 990.

Hospital Confinements	% Coagulase-positive Staphylococci Resistant to					Total Coag.-positive Staphylococci
	Sulphonamide	Chlortetracycline	Chloramphenicol	Penicillin	Streptomycin	
Hospital						
W	19·3	3·3	7·9	74·7	8·2	367
E	17·7	3·1	8·1	65·7	9·9	323
M	30·8	6·4	1·8	76·4	6·4	110
Total	20·2	3·6	7·1	71·3	8·6	800
Home Confinements	23·5	0	0	23·5	11·8	17

The nasal discharge which is nearly always present will provide a suitable specimen from which an assessment of antibiotic sensitivity can be made. Specimens should also be taken from any collection of pus requiring drainage. Even the bloodstained discharge drained from Case IV, although not apparently purulent, contained the staphylococcus. Nor should it be forgotten that this is a septicæmia, and that organisms may be cultured from the blood. Had such a specimen been taken from Case VI when it was first seen a delay of twenty-four hours might have been avoided. Such a delay may materially affect the chance of survival from the septicæmia and the choice of erythromycin in Case VI, before the sensitivity report was available, can only be regarded as a fortunate coincidence, for the sensitivity of a particular strain of staphylococcus is quite unpredictable.

Unless there is some contra-indication it is suggested that therapy should begin with penicillin, for at least some cases will respond to it. The drug should be given by the intramuscular route, for the effective dose is known then more certainly than would be the case with oral administration. Using the latter method, unexpected vomiting would reduce the effective dosage, and one is at the mercy of intestinal absorption rates. Dosage should be liberal, for it is better to err on the side of giving too much antibiotic, and there is no harm in giving 500,000 units of crystalline penicillin every six hours.

Unless the child is gravely ill when first seen, or does not improve in the first twenty-four hours, or there is significant deterioration during that time, therapy should be continued unchanged until the sensitivity report is available. Therapy may then be altered, but clinical progress rather than the result of an *in vitro* test should be relied upon in making the decision.

If deterioration is apparent before the sensitivity report is available, alternative therapy may have to be used, and this must inevitably depend upon guesswork, however intelligently it may be applied. The range of antibiotics is wide, and is continually increasing. Using the argument that the agents which have been discovered most recently are those to which the staphylococcus is least likely to be resistant, the choice would seem to lie between erythromycin and novobiocin.

Antibiotic therapy should not be terminated until the primary abscess has resolved completely. Again, it is better to err towards too much treatment. One relapse in Case V probably caused by the premature cessation of antibiotic therapy.

Operative procedures at this stage should be restricted to the minimum. These small patients are in no state to tolerate prolonged anæsthesia or major procedures, and the methods of antrostomy and counter-drainage from the orbit into the nose which Asherson found necessary have no place in therapy today. Trueta and Morgan (1954) have suggested that

Commentary. This child was by far the most gravely ill of the whole series, and, but for the successful "blind" use of erythromycin, would probably have died from the initial septicæmia. The continuing sensitivity of the staphylococcus to erythromycin, in circumstances where resistance should in theory have developed, is noteworthy. This is the only example in the series where any deformity (ectropion) has resulted from the infection. Even so, the correction of the ectropion, if it is necessary, should present little difficulty.

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TABLE II

Author	Case No	Age	Treatment	Duration	Result
Jacoby and Sagorin	1	3/52	Penicillin	11 days	Recovery
	2	6/52	Penicillin	10 days	do
Dunphy and Frazer	1	9 days	Penicillin Sulfathiazole	4 weeks	do
Howarth	1	4/52	Penicillin, Sulfathiazole, Drainage	6 weeks	do
	2	4/52	Penicillin, Drainage	2 weeks	do
	3	7/52	Penicillin, Drainage	4 weeks	do
	4	12/52	Penicillin, Sulfadiazine, Drainage	18 days	do
	5	15/52	Penicillin, Sulfadiazine, Drainage	30 days	Death (acute mastoiditis and gastroenteritis as complications)
Howard and Robinson	1	2/52	Penicillin	15 days	Recovery
Cohen	1	3/52	Penicillin	1 month	do
Macbeth	1	6/52	Penicillin, Drainage	Not known	do
McCash and Rowe	1	4/52	Streptomycin, Drainage	3 months	do
	2	4/52	Streptomycin, Chloromycetin, Aureomycin, Drainage	3 months	do
Reid, Spiro and Wilson	1	18 days	Sulfatriad, Penicillin, Chloromycetin, Aureomycin, Incision	1 month	do
	2	6/52	Chloromycetin	1 month	do
	3	1 year	Aureomycin	5 weeks	do
	4	3/52	Penicillin, Sulfamezathine, Terramycin, Incision	5 weeks	do
Gregson	1	5/52	Penicillin, Aureomycin, Drainage	2 weeks	do
	2	6/52	Penicillin, Streptomycin, Penicillin (repeated)	18 months	do (Sequestrum extruded)
	3	3/52	Penicillin, Aureomycin	3 weeks	do
	4	3/52	Penicillin, Streptomycin, Drainage	2 weeks	do
	5	1/12	Terramycin, Penicillin (repeated), Drainage	9 months	do
Gregson and Yeoman	1 (6)	15/12	Erythromycin (repeated) Drainage	9 months	do (Sequestrum extruded)

Summary

1. Six cases of acute osteomyelitis of the maxilla in infants are presented. There was no death from the disease, but three cases suffered one or more recrudescences of infection.

2. The pathology of the condition is discussed. The disease is thought to be a primary septicæmia with secondary embolic manifestations in the bone of the maxilla. Reasons are advanced to explain the propensity of the maxilla to act as a nidus for emboli at this particular age.

3. The changing pattern of therapy is discussed, and a policy for the use of antibiotics is suggested.

4. Methods of treatment by surgery are considered. Operative measures in the early stages should be limited to drainage of pus or decompression of the orbit by simple incisions. In the later stages exploration for sequestra may have to be undertaken, but reasons are advanced for delaying this measure as long as possible.

5. The change in prognosis since the introduction of antibiotic therapy is considered. A mortality of 25 per cent. in 1939 has been reduced to less than 5 per cent. during the last decade.

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THE MOVEMENTS OF THE LATERAL WALLS OF THE NASOPHARYNX: A CINE-RADIOGRAPHIC STUDY

By R ASTLEY (Birmingham)

MOST cine-radiographic studies of the pharynx have been concerned with movements visible in the lateral view Moreover, they have been concerned mainly with the oropharynx, the nasopharynx and particularly its lateral walls have received little attention To remedy this omission it was decided to attempt cine-radiography of the nasopharynx in a frontal projection

Method

The subjects were four healthy medical student volunteers aged 19 to 21 years The nasopharynx was coated with radio opaque material as follows with the subject supine, the neck was extended over a pillow and the head turned to one side Then about 2 mls of a 100 per cent weight in volume barium sulphate suspension ("Micropaque ') was injected through a short piece of fine plastic tubing inserted an inch or so within the lower nostril After sniffing and swallowing several times, the subject turned to the opposite side and a similar injection was made into the other nostril

Cine radiography was performed by the method described previously (Astley and Oldham, 1956), using an image intensifier and a 16 mm cine camera This technique employs intermittent radiation, so that each cine frame is exposed as an individual high speed (0 0025 secs exposure) radiograph, thus reducing movement blurring Films were taken at 16 frames per second in a supine postero-anterior position (the X-ray tube below the subject), with the central rays passing along the saggital axis of the hard palate (i e with the chin slightly raised so that the radiographic base line was tilted about 10 degrees above the vertical) The exact positioning was controlled by fluoroscopy through the image intensifier In addition, cine films of each subject were also taken in the lateral position

The cine-radiographs were examined as continuous loops, using a Specto analysing projector, when required the film could be slowed down or stopped to study any individual frame. The image was projected through a prism on to a horizontal surface, at life size, so that tracings could be made.

Results

In all four subjects the lateral walls of the nasopharynx could be distinguished and their movements observed. In two subjects the demonstration was particularly good.

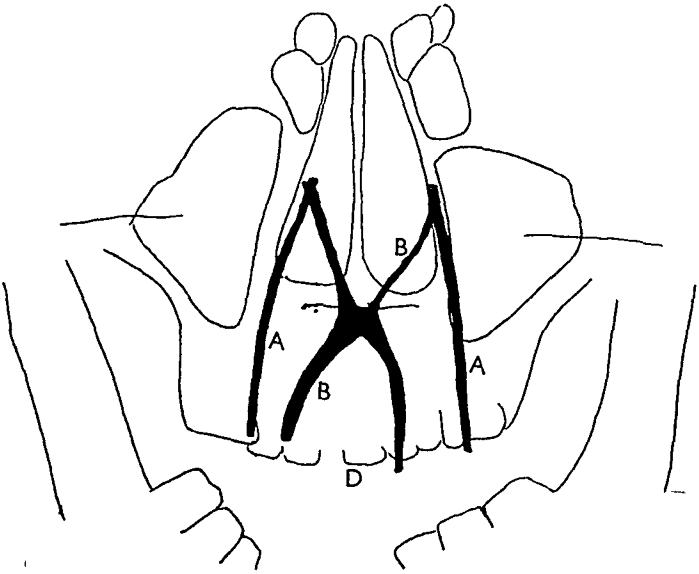

FIG. 1.

Normal female, age 21 years. Tracing from cine film.

A represents barium on the lateral pharyngeal walls during quiet respiration.
B shows their position when phonating "ah".
D is the level of the crowns of the upper teeth.

Quiet respiration. The walls showed as two thin, slightly curved white lines of barium. Their upper parts converged slightly and approximately overlay the shadows of the lateral walls of the nasal cavity (Fig. 1A). No movements were seen.

Deep respiration. There was slight outwards movement of the middle parts of the white lines, with a slight increase in their widest separation.

Phonating "ah". The lateral walls showed a pronounced inwards shift, maximum at about the level of the palate, where they touched together (Fig. 1B). The slight medial concavity of the white lines was replaced by a greater lateral concavity so that their combined shape resembled that of an hour-glass.

Blowing against the closed lips There was a similar inwards shift but the approximation of the walls extended over a longer segment The barium was squeezed out of this region, an indication of the firmness of the closure of the lumen (Fig 2)

Speech This was accompanied by to and-fro movements maximum at about the level of the palate

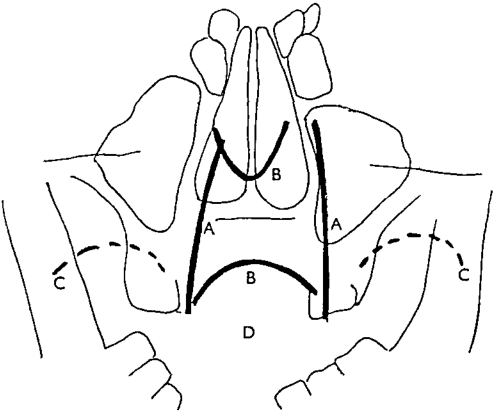

Fig 2

The same subject

A is the position of the walls during quiet respiration
B is their position when blowing against the closed lips the barium is squeezed out of the area of apposition
C is the upper margin of air in the ballooned cheeks
D is the line of the crowns of the upper teeth

Discussion

Comparison of the disposition of the barium in the frontal and lateral pictures left no doubt that it was coating the nasopharyngeal walls It also extended downwards to a slightly lower level and the lower parts of the two white lines below the level of the palate as seen in the frontal view appeared to represent the lateral walls of the oropharynx The region of maximum inwards movement and approximation of the white lines corresponded to the area of closure seen in the lateral view where the soft-palate was raised against the forwards bulging posterior wall of the nasopharynx

Thus it seems certain that the mechanism of closure of the nasopharynx is not confined to the events that are observed in the lateral picture. It is evident that the lateral walls of the nasopharynx play an important part by virtue of their considerable movement.

I am indebted to Mr. W. Stirk Adams, F.R.C.S, who suggested this study.

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CLINICAL RECORDS

BILATERAL LUXATION OF THE INCUDO-STAPEDIAL JOINT

By LENNART GISSELSSON (Orebro Sweden)*

In the October number of this Journal of 1956 Hallpike and Harrison asked whether localized freeing of the stapes should have been preferred to a fenestration procedure in a case with bony fixation of the stapes and, if so, all fenestration operations should begin with a diagnostic examination of the stapes at points other than its footplate I should like to answer both questions in the affirmative and even go so far as to state that in all cases of conducting hearing impairment that cannot be explained by examination of the drum, the middle ear should be explored Rosen has described a simple method for exposing the middle ear Since the procedure can be used without damage to the drum or any part of the middle ear and since it provides a good view

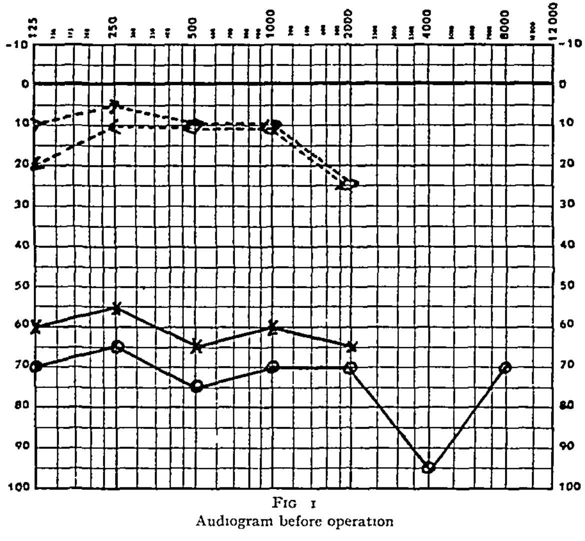

Fig 1
Audiogram before operation

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* From the Aural Department Central Hospital Orebro

of the sound conducting organs, it may be applied without hesitation for surgical exploration of the middle ear.

The justification of this attitude is substantiated by the improvement of hearing achieved in the following case.

A male, now aged 33, had been in a train accident eight years ago and sustained injury to the head. He was unconscious for two days. On return of consciousness he noticed that hearing was impaired on both sides. He reported that hearing capacity had been normal before the accident. Examination then suggested the hearing loss to be of neural origin. A year or so later, however, it was obvious that reduction in hearing was due to a conductive lesion. Operation was not considered and the patient was given a hearing aid, with which he managed quite well. Last year, eight years after the accident, the patient had to be examined for assessment of insurance claims. Both drums were found to be normal. The audiogram showed a conductive hearing impairment, but with slight neural involvement (Fig. 1). In an attempt to trace the cause of the impairment it was decided to explore the middle ear on the right side. The long process of the incus was found not to be in contact with, but to terminate a few millimetres short of, the head of the stapes. When attempting to replace the long process of the incus, the latter was found to be quite soft and atrophic. Since it was not possible to bring it into contact with the stapes, the body of the incus was pushed down between the head of the stapes and the

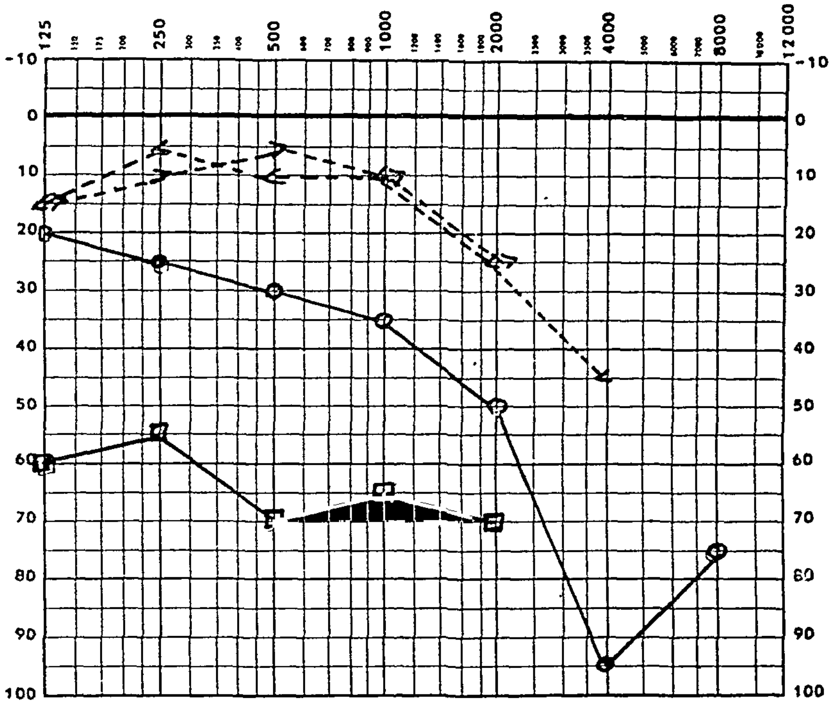

Fig. 2.
Audiogram 2 months after operation on right ear.

drum The drum was then pressed against the stapes with a gauze plug The operation resulted in a marked improvement in hearing (Fig 2)

In view of this success it was decided to operate on the other ear, too Operation, two months later, revealed that the long process of the incus was not in contact with the stapes on that side either It was situated behind the head of the stapes and terminated at the level of the stapedius tendon The long process was not atrophic, it was lifted and replaced Exact reposition was not possible the long process terminated a few millimetres short of the head of the stapes An audiogram recorded during operation showed such good improvement in hearing that it was decided not to try to better the anatomical relationship between the long process of the incus and the head of the stapes

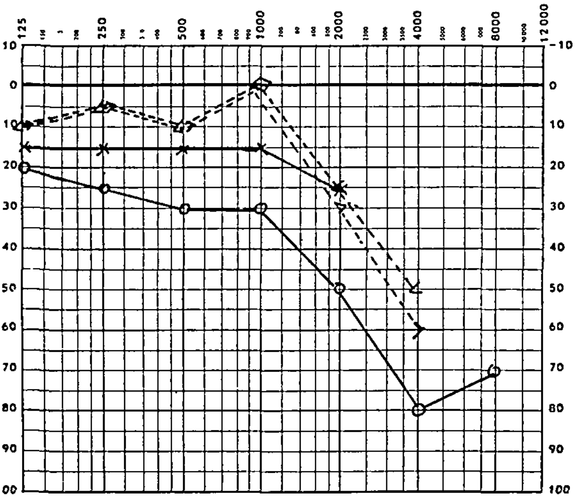

Fig 3
Audiogram 1 month after operation on left ear

After operation, hearing was so good that the patient no longer used his hearing aid An audiogram recorded one month after the last operation is given in Fig 3

In cases of conductive hearing impairment without demonstrable drum changes the next step in the diagnostic examination should be exploration of the middle ear The findings then made dictate what therapeutic measures should be taken

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INFECTIOUS MONONUCLEOSIS REQUIRING SPLENECTOMY AND TRACHEOTOMY

By A. S. DAVIDSON (Kettering)

DURING the recent epidemic of "Asian influenza", a young unmarried woman aged 22 was admitted as an emergency in a semi-collapsed condition.

She had apparently been in good health until a fortnight previously, when she developed "'flu", i.e. sore throat, headache, fever, rhinorrhœa and cough. Initially she retired to bed and had a course of oral penicillin. After three days she felt better and got up, but only to feel worse, and then to return to bed. Because of her continuing fever and sore throat, she then had a course of chloromycetin and later terramycin. Despite this treatment, however, her general condition deteriorated and the sore throat became very much worse in the two days prior to her admission. In addition she had difficulty in breathing through the nose, and spoke with a nasal intonation.

At about 9 a.m. on the day of admission she experienced severe abdominal pain and almost fainted. The pain did not abate, and after an interval she noted pain in the left shoulder. Shortly afterwards she had a somewhat loose motion. There had been no urinary symptoms, and she had just concluded a normal trual period. She was collapsed and seriously ill, and her general prac- ner telephoned the Ear, Nose and Throat Consultant requesting a domici- consultation. As the patient lived in an outlying area, and it was obvious that she was dangerously ill, it was deemed safer to admit her immediately to the Ear, Nose and Throat Department for assessment.

On examination in the ward at about 11 a.m., the most striking feature was her general collapse and pallor. Blood pressure was 80/50, temperature 99·8, pulse 136. Scrutiny of the throat was not very rewarding. There was a patchy opaque whitish film on the posterior pharyngeal wall superiorly, but the tonsils were not noticeably abnormal. Bilateral slight enlargement of the lymph glands in the superior deep cervical group was noted, but no glandular enlargement or tenderness was present in the posterior cervical triangles, axillae, or groins. Abdominal examination did not reveal any definite localizing signs, and as the nature of the abdominal catastrophe was still not clear, guidance was sought from the General Surgeon.

Shortly after noon she was seen by my surgical colleague, Mr. T. H. Cullen.

Radiating pain now involved both shoulders. Abdominal examination showed loss of liver dullness, and dullness was present in both flanks. There was maximal tenderness and some guarding across the upper abdomen, with slight tenderness in the lower abdomen, and marked tenderness on rectal examination. Moving the patient on to the left side caused intense pain in the left abdomen. The blood picture was as follows: Hæmoglobin=80 per cent., Serum Amylase 200 units per cent , W B C 19,400 per c mm , of which 53 per cent were Neutrophils, 42 per cent Lymphocytes, and 5 per cent Monocytes, with a few atypical Lymphocytes

The possibility of acute pancreatitis had been borne in mind, but with this suggestive blood picture a diagnosis of infectious mononucleosis, with the rare complication of splenic rupture was thought likely, and laparotomy was advised

The operation was carried out at about 2 p m , and the abdominal cavity was found to be full of clot and dark blood The spleen was grossly enlarged with a subcapsular hæmatoma, and there was active bleeding through a tear in the capsule Splenectomy was performed Blood transfusion was started at the time of operation and in all, three pints were given over the next six hours

In the case of this patient with severe collapse, following a toxic illness of some weeks, and known to have a throat infection the risks of anæsthesia were not inconsiderable and careful thought was given to the agents used Cyclopropane and oxygen was thought to be the least disturbing mode of induction Upon its administration, however, as soon as there was the slightest diminution of consciousness, great difficulty was experienced in achieving an adequate airway By constantly keeping the tongue well forward, and after twice interrupting the progress of anæsthesia because of cyanosis, it finally became feasible to administer some Flaxedil, and to insert an endotracheal tube through the mouth at which stage maintenance of anæsthesia became relatively straightforward It was anticipated that there would be some difficulty in maintaining an airway post operatively, on removal of the endotracheal tube and the E N T Consultant was accordingly requested to be in attendance

The opportunity was taken to examine the pharynx and larynx at leisure, and with all the necessary facilities to hand It was seen that the milky pharyngeal exudate was continuous with a large pale fibrinous clot filling the post-nasal space and just visible below the soft palate A gentle manœuvre with the pharyngeal sucker completely loosened the mass easily and without bleeding On examination the appearance of the larynx was normal but the pharynx showed marked hypertrophy of the lateral pharyngeal bands commencing an inch below the tonsils, extending downwards to the pyriform fossae, and covered by a milky white exudate With the aid of the pharyngeal sucker, this exudate was also easily removed The lymphoid inflammation and hyperplasia at this level explained the severe and painful dysphagia, although on previous examination of the pharynx with a tongue depressor, there had been no noticeable lesion

It was thought now that the asphyxial episode during the induction of anæsthesia might have been caused by the airway having become blocked by a detached portion of the fibrinous post nasal mass, and it was decided to remove the endotracheal tube, but to detain the patient in the theatre until it was quite clear that there was no risk entailed in returning her to the ward

When the pharyngeal reflexes had become very definitely established, the endotracheal tube was removed, and for five minutes the patient s condition was satisfactory She then became cyanosed, and on examination of the pharynx it was obvious that the obstruction was due to the hypertrophied lateral bands of lymphoid tissue falling inwards on to the tongue base with occlusion of the airway. Forcibly pulling forward the tongue, and administration of oxygen, relieved the condition temporarily. Finally, even when fully conscious, and with a stitch anchoring the tongue forward, the patient became cyanosed on withdrawal of oxygen. An oral endotracheal tube was therefore hastily replaced, and a leisurely tracheotomy performed under local anæsthesia and a nitrous oxide, oxygen mixture.

Apart from some crepitations at both lung bases, which persisted for the first five days after operation, recovery was uneventful with the aid of penicillin, and a later course of tetracycline.

One week after operation dysphagia had disappeared, and the pharynx showed a normal appearance on indirect examination; the tracheotomy tube was thereupon removed. There were no untoward abdominal occurrences, and during the next three weeks her temperature gradually subsided. She was then discharged for a period of convalescence.

Pathological Investigations

The diagnosis of mononucleosis was confirmed by a Paul Bunnell positive 1 : 320 on the day following operation, and positive 1 : 80 on the twentieth day.

Culture from a throat swab taken on admission grew micrococci with a few colonies of *N. catarrhalis*. No K.L.B. were isolated, and no Vincent's organisms were seen.

The spleen was enlarged to about three times its normal size, and on section the appearance was compatible with a diagnosis of mononucleosis.

Section of the post-nasal mass showed it to consist of a mucoid-fibrinous exudate, infiltrated chiefly by lymphocytes, and some atypical monocytes.

Discussion

Attlee in 1932 first recorded spontaneous rupture of the spleen, in what was inferred from his findings to be a case of mononucleosis. In a review of the subject in 1952, Belton tabulates at least eighteen further cases, and an additional case under his care successfully treated by operation.

It would appear probable that the spleen is commonly enlarged in approximately the third or fourth week of the illness (Smith and Custer, 1948), and that the trabeculae and capsule become infiltrated to a varying degree with normal and abnormal lymphocytes. The enormous increase in size of the spleen would seem to lead to a tear in the weakened capsule, either spontaneously whilst asleep, for example, or following an additional increase in splenic congestion such as might occur during defæcation. In Belton's case, rupture was associated with the muscular activity related to digging (Belton, 1952).

In some instances the hæmorrhage spreads in the subcapsular plane, and for a time remains localized, only to rupture into the general peritoneal cavity at a later stage, with exacerbation of pain, and evidence of internal hæmorrhage such as pallor and collapse. As the amount of blood in the peritoneal cavity increases, the classical signs of shifting dullness may be manifested. Severe abdominal pain appears to be rare in uncomplicated mononucleosis, and in one such series of 81 cases, only one had abdominal pain, and that of a mild nature (Hoagland and Gill, 1955). In another series of personally observed cases only two out of a total of 153 had severe abdominal pain; one of these had a ruptured spleen, but in the other case operation showed the spleen to be intact (Hoagland and Henson, 1957). More difficult to recognize are those instances where bleeding is massive from the start. Ziegler and Stobbe each record a fatal case where there is no mention of abdominal pain. Ziegler's patient felt weak at 10.30 a.m. and was dead at 1.30 p.m., whilst the young man reported by Stobbe suddenly felt extremely weak and was dead twenty minutes later.

When a patient known to have had a sore throat for a period of weeks experiences severe abdominal pain, or severe and sudden collapse, the likelihood of mononucleosis and splenic rupture is therefore well borne in mind.

When considering complications of infectious mononucleosis, references to respiratory obstruction in the literature occur less frequently than do references to splenic rupture. In a review of the subject in 1955, records of eight cases necessitating tracheotomy are listed (Mandel, Marilley and Gaines). So far, the writer has not encountered the record of a case of mononucleosis in which both complications were encountered in the same patient.

In this particular patient the question arises as to the chain of events leading up to the sudden deficiency in the airway following splenectomy. It would be possible to attempt an explanation in terms of œdema of the glottis aggravated by an anæsthetic requiring intubation. The appearances on direct laryngoscopy as detailed above, render such an explanation both untenable and unnecessary, and lend weight to the respiratory obstruction encountered by the anæsthetist prior to intubation. When in retrospect the history was recalled, it was clear that for several days prior to her admission, dysphagia had greatly increased, and it is likely that even then her pharyngeal airway was only just adequate. Rupture of the spleen, and the consequent copious bleeding into the peritoneum, produced a profound state of collapse. In this critical state the total expenditure of energy became minimal, so that the respiratory exchange appropriate to this state of diminished vitality was less than that existing before the catastrophe. This basic respiratory exchange at a lower level, was capable of being maintained through the restricted pharyngeal lumen, and in this manner it seems plausible to argue that the disaster of splenic rupture, staved off the arrival of respiratory obstruction.

Following her successful resuscitation by splenectomy and blood transfusion, with restoration of normal blood pressure, the metabolic demands increased. The necessary gaseous exchange was now greater, and the latent inadequacy of the airway therefore became apparent, even allowing for the fact that by now the patient was fully co-operative, and had full use of the accessory respiratory muscles.

Reports of findings in the post-nasal space in infectious mononucleosis are rare, and the writer was only able to trace one. This occurred in a comprehensive review of the pathology of mononucleosis by Custer and Smith (1948), in which they analyse the autopsy details of nine fatal cases. In one instance the cause of death was hæmorrhage of the naso-pharynx, but unfortunately no material from the nose or naso-pharynx was available. It is likely that the essential lesion is lymphoid hyperplasia, and that in some instances such as that reported by the writer, massive exudation occurs consisting of a mucoid fibrinous matrix heavily infiltrated by lymphoid cells. In other instances areas of necrosis may develop, giving rise to massive hæmorrhage, as in the fatal case recorded.

In a patient seriously ill from anginose mononucleosis, detailed examination of the nose and naso-pharynx is likely to be overlooked. The likelihood of a lesion therein is, however, of more than academic interest, because in some patients with threatened respiratory obstruction due to massive tonsillar hyperplasia, it has been recommended that the use of a temporary nasal tube may avert the need for tracheotomy. This may be so, but the risks of serious nasal or post-nasal hæmorrhage must be taken into consideration.

Cortisone

Corticotrophin (A.C.T.H.) certainly seems to offer great possibilities in the treatment of mononucleosis. There is a very informative account with a list of four other articles dealing with this special point, in the paper by Mandel, Marilley, and Gaines (1955) previously mentioned. In their case with the threat of possible respiratory obstruction, œdema of the pharynx and larynx was a noticeable feature, and the administration of A.C.T.H. averted the necessity of tracheotomy. In the writer's patient the lateral bands of lymphoid tissue appeared hyperplastic and inflamed, rather than œdematous, and without tracheotomy it seemed impossible to maintain the airway for sufficient length of time to try the effects of A.C.T.H. A further consideration was that administration of A.C.T.H. was thought likely to interfere with the processes of healing following the abdominal operation. Nelson and Delaney (1953) describe a proven case with gross tonsillar enlargement and with exudation, which was treated with Terramycin and Cortisone for 24 hours. Impending respiratory obstruction worsened, however, and tracheotomy was performed with a successful outcome.

In reviewing the treatment of cases which occur so rarely, and in which the respiratory obstruction may occur at different anatomical levels, e.g. fauces, pharynx or larynx, and from different causes, e.g. œdema, exudate or lymphoid hypertrophy, it is evident that each individual case must be considered on its own merits. If time can be bought with Corticotrophin, tracheotomy may be averted. Should this not be possible, then the relatively minor operation of tracheotomy must be carried out.

Owing to the extremely varied appearances seen on examination of the pharynx, it is very likely that some fatal cases of anginose mononucleosis have in the past thought to be swab negative diphtheria, or even an acutely developing reticulosis, or "atypical" lymphosarcoma, since infectious mononucleosis is commonly thought of as essentially a benign disease. An awareness of the more severe and bizarre forms of the disease may, however, lead to a revision of this opinion and to the consequent earlier elucidation of the disease in its more obscure manifestations.

Acknowledgments

I gratefully acknowledge my indebtedness to my colleague, Mr T. H Cullen, who advised, and personally undertook the operation of splenectomy, and also to Dr. H N. S Wilkinson, who overcame the hazards of a most difficult anæsthetic problem.

My thanks are due to Dr H J. Voss, for his helpful advice concerning the pathological findings

I wish to thank those members of the Nursing and Resident Medical Staffs who did so much for this patient, and gladly acknowledge that without their skill and care, the successful outcome could not have been achieved

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SOCIETIES' PROCEEDINGS

ROYAL SOCIETY OF MEDICINE—SECTION OF OTOLOGY

President: I. SIMSON HALL, F.R.C.P.(Ed.), F.R.C.S.(Ed.)

December 6th, 1957

Discussion on Stapes Mobilization

THE opening papers by Dr. Rosen, Mr. Terence Cawthorne and Mr. Venters will be found on pp. 263-291.

MR. A. FINLAY BROWN. Over the past year, in a small series of 31 cases (34 ears), there were 13 males (14 ears) and 18 females (20 ears). Failures numbered 7 males (50 per cent.) and 5 females (25 per cent.), was this predominance of failures in the male also found by other surgeons?

In six cases the footplate was attacked. Three of those were fenestrated according to Dr. Rosen's technique; one at the superior margin, one in the centre of the footplate and a third at the inferior margin. The immediate results were very satisfactory, but in 24 hours the hearing had returned to the preoperative level, and remained there ever since. In the remaining three cases after both the crura had been fractured using the indirect method, the footplate was also fractured. The results in these cases were a complete closure of the air/bone gap. The improvement had been maintained for eight months in one case, and three months in another; the third case, also improved, was too recent to warrant its inclusion. Is there an explanation for the failure of the fenestration of the footplate, and what are the experiences of other surgeons using this fenestration technique?

MR. S. W. ALLINSON said that in his opinion the stapedius muscle tilts the stapes back and out and acting with the tensor tympani helps to keep the joint surfaces of the ossicular chain in contact. These muscles also keep the stapes in a suitable position and in readiness to receive sound waves.

Thus it is not sufficient to mobilize the stapes if the best results are to be obtained, but the anterior part should be mobilized and the delicate posterior part of the footplate and adjacent margin of the oval window should be preserved as much as possible so as to enable the stapedius muscle and tensor tympani muscle to continue to function normally.

MR. H. D. BROWN KELLY said that he would like to add to the welcome given to Dr. Rosen. He had had the pleasure of watching Dr. Rosen operate on his visit to Glasgow and in his clinic in New York, and was very grateful for his teaching. With regard to the fenestration of the footplate, what happened to the hole? It was not covered by any flap as in a canal fenestration, and it presumably filled with granulation tissue Had Dr Rosen had the opportunity of examining a footplate fenestration some months after it was performed?

MR WILLIAM McKENZIE asked Dr Rosen about the possibility of adhesions forming in the middle ear as a result of the operation He had found these several times, and most often in young people They seemed to be the result of hæmorrhage rather than of infection, and hæmorrhage in the middle ear after operation could not be controlled

These adhesions might determine the number of times the ear could be explored

THE SCOTTISH OTOLARYNGOLOGICAL SOCIETY

THE seventy-fifth meeting of the Society was held in the Royal Infirmary, Glasgow, on Saturday, November 23rd, 1957 The President, Dr E A M Connal was in the chair *Cases were presented by the staff of the Royal Infirmary* E N T Department, Messrs Stephen Young, A R Harper, J R McCallum, T Cartmill, and J T Lees

Ten cases of nasal sinusitis in children treated by radical operation were demonstrated and discussed Patients with malignant disease of pharynx and larynx treated by various methods, surgical and radiological were also shown Other interesting cases included ectopic thyroid tumour in the trachea, eosinophil granuloma of the skull simulating an acute mastoid, three cases of dysphagia lusoria, and a tracheal reconstruction without the use of grafts Dr W G Hood showed a case of otosclerosis treated by bilateral stapes mobilization, with a very good result

In the discussion which followed, the advisability of radical antral operations on cases of mixed infection and allergy was strongly questioned by Mr Gavin Young, Mr Collins, and other members Dr Birrell described the intra-antral injection of hydrocortisone in a series of cases with encouraging results Mr Brown Kelly suggested that many nasal and aural troubles in children were kept up by repeated infection from parents harbouring streptococci in tonsils or sinuses Mr Land stressed the role of the over-anxious parent in allergic conditions in children

Dr Morrison, in describing the radiological treatment of malignant disease in ear, *nose and throat work mentioned the variety of tissues to be dealt with* Tumours involving bone (e g antrum) were unsatisfactory from the radio therapy viewpoint In the larynx, the tumour is often isolated from normal tissue, and in these cases surgery is advised Dr Glennie described the routine tomograph in the assessment of regression in the laryngeal tumours He recorded several successful cases treated by radium implants

GENERAL NOTES

The Editors are anxious to improve the clinical aspect of the Journal and would welcome more reports of interesting cases. These clinical records need not be at all elaborate and it would be unnecessary to go minutely into the ætiology or the literature of the subject. Senior Registrars have unique opportunities for seeing a large variety of cases, many with unusual features. It is in many ways regrettable that these should go unrecorded. It is hoped that all those in charge of clinics will help us in this matter and see that such cases are not overlooked.

OTOLARYNGOLOGY LECTURES 1957-58

The following lectures, arranged jointly by the Royal College of Surgeons of England and the Institute of Laryngology and Otology, will be delivered in the Lecture Hall of the College, at 5.30 p.m.

1958
May 1st. Professor W. D. M. Paton. The pharmacology of mucous membrane.
July 3rd. Mr. Norman C. Tanner. Haemorrhage from the Œsophagus.

INTERNATIONAL CONGRESS ON THE MODERN EDUCATIONAL TREATMENT OF DEAFNESS

An International Congress on the Modern Educational Treatment of Deafness will be held at the University of Manchester from July 15th-23rd, 1958.

The Congress is being organized with the approval and support of the Council of the University, with financial help from the Ministry of Education, the National College of Teachers of the Deaf, and the Rathbone Trustees, and with the co-operation of the British Council and the Alexander Graham Bell Association for the Deaf, Inc., Washington, D.C., U.S.A.

The Congress is being planned to bring together educators, otolaryngologists, pædiatricians, medical officers, audiologists and educational psychologists, to consider and discuss the significance, to the future development of their work, of the results of modern research on problems of deafness in childhood.

Arrangements are being made to offer accommodation to members of the Congress in University Halls of Residence. The cost of this type of accommodation is expected to be about £1 2s. per day, including bed, breakfast and dinner. The University Refectory will be available for meals during each day.

Forms of application for membership of the Congress will be supplied upon request, to Professor A. W. G. Ewing, Department of Education of the Deaf, the University, Manchester 13, England.

General Notes

THE BELGIAN SOCIETY OF OTOLOGY, RHINOLOGY AND LARYNGOLOGY

From 2nd to 7th June, 1958, an Extraordinary Congress, under the Patronage of the Commissariat General of the Belgian Government will be held at Brussels, at the Universal and International Exhibition, Brussels, 1958 President Prof J van den Branden (Brussels)

SCIENTIFIC PROGRAM

(1) Cancer of Larynx First European meeting of the International Commission for the Study of the Cancer of the Larynx, (2) Functional Surgery of the Ear, (3) Meeting of the Sociéte de broncho œsophagologie de Langue Française, (4) Meeting of the International College of Experimental Phonology, (5) Festival of *scientific film in E N T field*

Every participant in the Congress will be granted an A1 priority concerning accommodation and free access to the exhibition from 1st to 8th June

The social program and all other informations will be communicated later

Inscription fees Effective members, Fb 500 Adherent members, Fb 250

For any information, kindly apply to the General Secretary, Dr Henschel C E L , 19 Rue de Fétinne, Liege, Belgium

FACULTÉ DE MÉDECINE ET DE PHARMACIE DE BORDEAUX

Clinique Oto-Rhino-Laryngologique (Annexe Saint Raphael, 3 Rue Jean Burquet) Cours de Perfectionnement du Professeur Portmann avec la collaboration de M le Professeur J Despons, de M le Professeur agrege Michel Portmann, de MM les Professeurs Aubertin, Bessiere, Broustet, Delmas Marsalet, Lachapele, Pautrizel, Piechaud, et de MM les Docteurs Berger (oto-rhino-laryngo logiste des Hôpitaux), H Pouyanne (neuro-chirurgien des Hopitaux), Robin (ancien chef de Clinique), G Lacoste (bronchoscopiste), J Pommez (phoniatre), J L Barbe (ancien chef de Clinique adjoint), J Rivasseau (allergiste) Lundi, 19 Mai au Samedi, 31 Mai, 1958

FACULTE DE MÉDECINE DE PARIS

Chaire de Clinique Oto Rhino Laryngologique Professeur A Aubin Cours de Technique Chirurgicale—La Chirurgie de la Surdite Lundi, 9 au Samedi 14 Juin, 1958

Apply Pavillon Isambert, Hôpital Lariboisiere, 2 Rue Ambroise Paré, Paris

General Notes

ROYAL SOCIETY OF MEDICINE

SECTIONS OF LARYNGOLOGY AND OTOLOGY

Summer Meeting in Edinburgh, June 27th, 28th, 1958.

Friday, June 27th.

9.30 a.m. Scientific Meeting: Royal College of Surgeons of Edinburgh, Nicholson Street. *Discussion:* "Acoustic Neuroma". Professor Norman Dott. Dr. C. S. Hallpike. Dr. D. Dix.

1 p.m. Luncheon for Members and Guests. Carlton Hotel, North Bridge. Price 15s. inclusive of wines and gratuities.

Afternoon. Garden Party by invitation of the Presidents.

7.15 for 7.45 p.m. Dinner in The Royal College of Surgeons of Edinburgh. Tickets Two guineas each inclusive of wines.

Saturday, June 28th.

9.30 a.m. Scientific Meeting: Royal College of Surgeons of Edinburgh. *Discussion:* "The dysphonias and the scope of speech therapy and their treatment". (Illustrated by recordings). Mr. Maxwell P. Ellis. Mr. L. Willmore.

12.30 p.m. Meeting ends.

For details of accommodation write to the Secretaries of the Sections.

Registration Fee of 10s. will be made for each Member to cover overhead expenses.

Members wishing to attend the Meeting should apply *as soon as possible.* Bookings at hotels must be made through the Royal Society of Medicine and *not* directly with the hotels. Accommodation is limited and early application should be made.

Applications should be accompanied by a cheque *made payable to Dr. J. F. Birrell* to cover the registration fee, lunch and Dinner tickets, but *not* for hotel accommodation.

N. L. CRABTREE, Honorary Secretary, Section of Laryngology.
LIONEL TAYLOR, Honorary Secretary, Section of Otology.

The Journal of
Laryngology and Otology

(*Founded in* 1887 *by* MORRELL MACKENZIE *and* NORRIS WOLFENDEN)

May 1958

PNEUMATIZATION OF THE MASTOID BONE

By MARCUS DIAMANT (Halmstad Sweden)

IN a recently published series of articles by Tumarkin (1957), a critical appraisal of my (Diamant, 1940, Dahlberg and Diamant, 1945, Diamant, 1949) detailed investigations on mastoid pneumatization was included Exception was taken to my conclusions regarding

(1) The variation in size of the mastoid air cell system
(2) The relationship between its final size and the various forms of otitis
(3) The relative importance of hereditary and environmental factors in its growth

At the outset it may be stated that the basic dispute is located in the area of biological cause and effect relationships

In agreement with previously held views Tumarkin maintains

That the presence of small mastoid air cell systems (unlike large ones) is always
(1) indicative of pathological growth, and
(2) due to otitis infections

In contradistinction to previously held views I maintain

(1) That no sizes of mastoid air cell systems indicate *per se* a pathological growth, on the contrary, irrespective of their being small, medium or large, all sizes indicate, as a rule, anatomical i e non pathological growth
(2) That, as a rule, small mastoid air cell systems are not due to otitis infections, on the contrary, they indicate a proneness of the corresponding ears to contract discharging middle ear diseases

For the purposes of this discussion I wish to suggest the following definitions:

(1) The *size* of the mastoid air cell system means its extension measured with a planimeter on the radiogram in lateral projection.

(2) The *anatomical* size means its growth determined by hereditary and non-pathological environmental factors.

(3) The *pathological* size means its growth similarly anatomically determined by hereditary and non-pathological environmental factors, but additionally affected by pathological factors representing a disease entity, thus partaking in the determination of the final size and causing a halting, diminishing or even enlarging effect.

I. The Critical Appraisal of my Investigations Selection Factors, Radiological and Planimetric Methods

No questions were raised by Tumarkin concerning the selection factors used for the various groups in my studies. This holds true for the groups representative of: (1) the general population (1940 study), and (2) the various forms of otitis (1940 study) as well as (3) the families and twins (1945 study).

Tumarkin states no major objection to the radiological and planimetric methods used in my studies, but he does mention one item which he considers a significant omission. He claims that I ought to have included the mastoid antrum and the tympanic cavity in the area of the mastoid air cell system measurement. He maintains that, since these two anatomical formations are never missing, I would not have obtained any zero value if both were included.

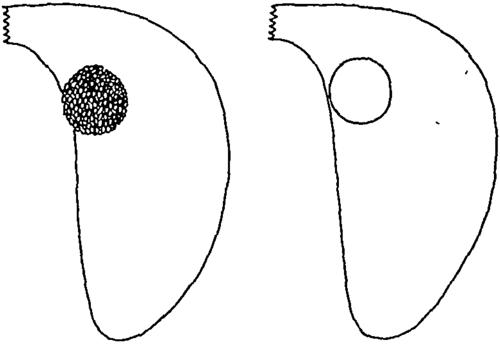
In pneumatized mastoids the area of the mastoid antrum is in fact included in the measurement, since it is covered by air cells in the lateral X-ray projection. In the acellular mastoid bone, however, the mastoid antrum is not covered by radiologically visible air cells. Thus, if Tumarkin's suggestions concerning this detail were followed, the *same* measurements would be obtained:

(1) When the air cell system is small enough so as to just cover the mastoid antrum.

(2) When the mastoid antrum is not covered by any air cells, i.e. in the acellular mastoid. The following schematic drawings show how the use of this method would lead to false values (Fig. 1).

Tumarkin further suggests that the tympanic cavity should also be included in the area measured, since it represents the air spaces in the temporal bone. My aim is to measure the size of the mastoid *air cell system*. Therefore, I do not include the tympanic cavity. Neither the tympanic cavity nor the mastoid antrum constitute mastoid *air cells*.

In my papers I have stressed the fact that the mastoid air cell system is not in itself claimed to be an influencing factor. Its size, measured with a planimeter on the radiogram in lateral projection, is used merely as an indicator valuable for deducing diagnostic and prognostic conclusions. Expanding the area measured by including the mastoid antrum twice or by adding the tympanic cavity would not change the logical sequence of my conclusions. Any studies measuring the same areas should be expected to produce similar results. Increasing the area of measurement, as Tumarkin recommends, would only introduce extraneous factors.



1 Mastoid antrum covered by air cells

2. Mastoid antrum not covered by air cells

FIG 1 Mastoid bone (*schematic drawing*)

Conformity with the Normal Curve Distribution

Tumarkin maintains that the frequency distribution of the various sizes in my age group of 10-30 years does not and cannot conform with the normal curve (Fig. 2). In his comments on my distribution curve showing the varying pneumatization in 178 individuals in this age group, Tumarkin writes: "Note that it includes cases with 'negative' pneumatization. Note also the skewness. These two points suggest that the curve is *not truly normal.*" In rebuttal to my conclusion that all sizes of air cell systems may be normal variants he stresses the fact that the range of variation, (1) actually goes down to size zero, which in normal variants he states is impossible, and, (2) statistically goes down below size zero, which he states is absurd.

First, the statement that the range of variation in normal variants cannot go down to size zero, is based on a teleological premise that the acellular mastoid must be pathological. According to teleological reasoning, every part of the body must have a function to justify its existence therefore, the fallacious corollary states, the acellular mastoid must be pathological. We will return to this question later. One other point needs to be stressed at this juncture. In any part of the human body many individual variations in the time of onset of anatomical growth are noted. For instance, some individuals show a distinct air cell system at birth, whilst in others the development has not as yet started. The distribution of the anatomical sizes, biologically varying at random, may be expected to conform to a normal curve already at the early phase of development. Consequently as a principle the size zero can be represented on the normal curve. Furthermore, the presence of air cells need not be visualized on the radiogram since they start developing around the walls of the mastoid antrum. Even visible air cells may be so minute that the measurement will show zero square centimetres, the standard error of the measurements exceeding 0·2 square centimetres.

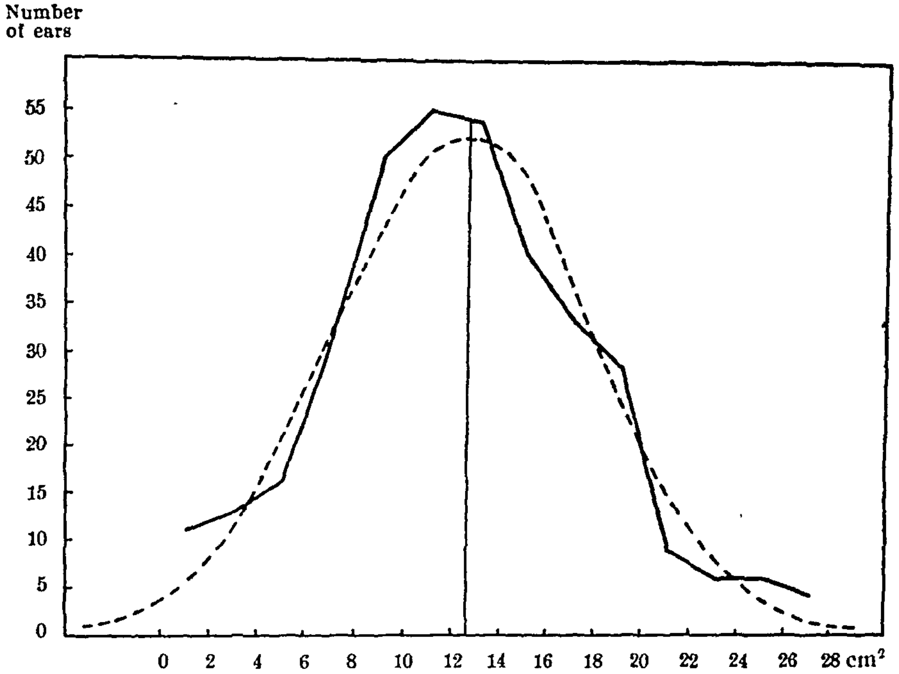


FIG. 2.

Distribution of the size of the cellular system (356 normal ears). The corresponding normal curve is also given (broken line) (Diamant, 1940).

Secondly, the possibility of having a distribution curve statistically going down below size zero obviously depends upon what is measured and what measuring method is used. In my investigation I measured visible air cells on the radiogram. Below size zero no air cells can be visible. The growth of the air cells, like all anatomical growth, must, however, be due to biological factors. I have found, together with Dahlberg, that the hereditary factors, computed singly, represent more that 84 per cent. of the total growth causative factors. The environmental factors, likewise computed singly, represent less than 54 per cent. of the total causative factors. Since the sum of these percentages exceeds 100, this in itself indicates that both hereditary and environmental factors are working to stimulate and to depress developmental growth. The clinical presence or absence of air cells is determined by the interaction of these two forces. The balance between these biological forces decides whether air cells will develop or not; the resultant falling above or below size zero on the distribution curve respectively. Thus, both the clinical presence and absence of air cells represents the interaction. With a positive resultant air cells are produced. With a negative resultant no air cells are produced. It could be suggested, for instance, that the biological factors with a negative resultant produce an increasing density of the mastoid bone. (It may be noted that microscopical slides from ears clinically free from middle-ear diseases are also free from all signs of previous or present inflammatory processes in the sclerotic mastoid bone.) A condensing effect cannot of course, be measured with a planimeter. But the fact that it cannot be measured with a planimeter does not eliminate my possibility of using a normal curve as a mathematical model. Neither does it substantiate the conclusion that anatomical factors do not operate nor that pathological factors do operate. The fact that the curve clinically goes down to size zero, and statistically goes down below size zero, is consequently not "absurd" as claimed by Tumarkin.

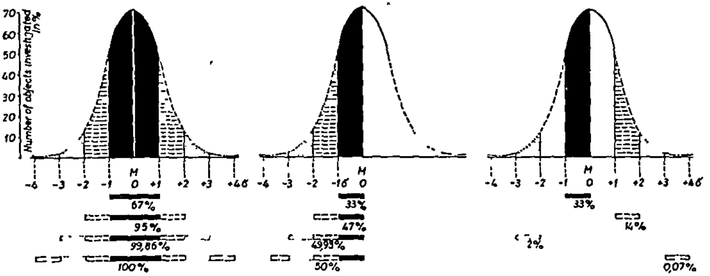


FIG 3

Variation in size or weight around the mean according to the computed value of σ, the mean is marked with 0

A further comment is in order regarding the vague expression "not truly normal". It must be stressed that an absolute conformity with a normal curve will hardly ever be found in biological size investigations. Fairly close conformity with a normal curve distribution was found in two separate samples, in my 1940 and 1945 studies. This dual conformity would seem to exclude coincidental findings.

Tumarkin's statement also observes that my distribution curve shows a skewness. This deviation was analysed in my 1940 monograph (page 65). There it was shown that although the statistical computations showed the presence of a slight skewness in the distribution curve, this skewness was small enough not to prevent the use of current formulae for the computation of standard error. According to Lindeberg the mathematical value of the skewness was—1·4 per cent.

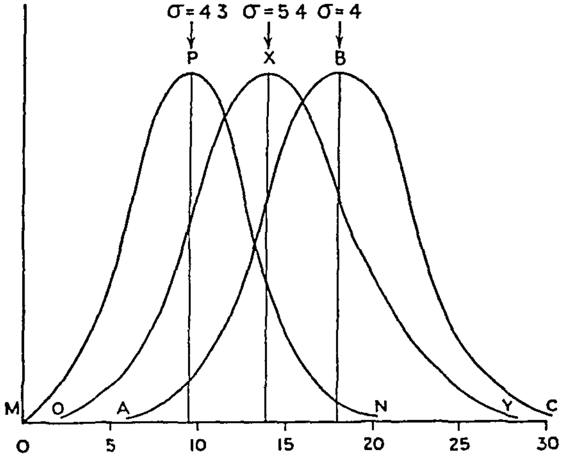


FIG. 4.

The significance of the *mean size*.

ABC is an ideal normal curve. OXY is a B curve shifted slightly to the left by a slight amount of disease. MPN is an A curve shifted greatly to the left by excessive disease.

The Significance of Deviating Curves

In size registrations of biological growth, the frequency distribution of the various size determinations in a large series representative of the general population may be expected to conform to a normal (probability) curve. The normal curve distribution can be seen in Fig. 3.

Both a shift to the left of the normal curve distribution and a skewed curve may be caused by differences in the age or sex distribution in the groups studied or they may be due to the selection factor chosen. Such deviations from the normal curve may be obtained when anatomical factors alone have influenced the growth as well as when pathological factors have superimposed their effect on the anatomical growth. When *a shift to the left* actually is caused by a pathological effect on growth, the pathological factors may be presumed to have exerted their influence fairly equally in *all* individuals or samples of the group. When pathological factors have exerted their influence on *a limited but appreciable section* of the group, this may be presumed to result *in a skew (or otherwise deviation) distribution curve.* This difference produced by influence on all or a part of the group respectively seems to have been disregarded in Tumarkin's study and will be reverted to later.

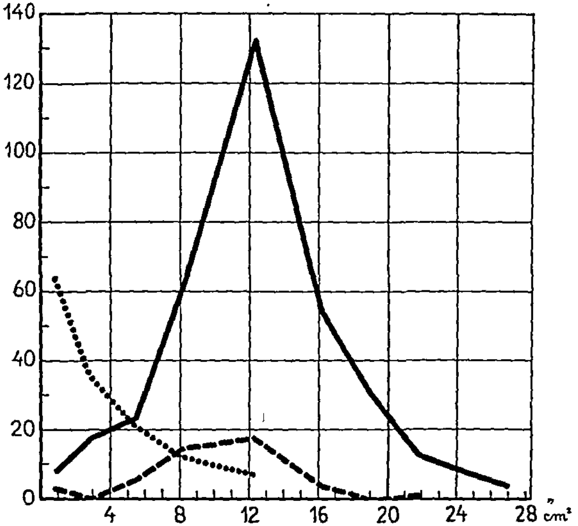


FIG 5

Normal material (solid curve), chronic otitis (dotted curve) and acute otitis (broken curve) at the ages of 10-30 years, grouped according to the size of the cellular system (Diamant 1940)

Tumarkin's statements given under the heading of "The significance of the mean size" (in his Fig. 16 on page 224 and my Fig. 4) indicate that he concludes that the shift to the left not only may, but in fact proves that a pathological influence on the growth of the mastoid air cell system has been operating. He claims (1) that disease has influenced, (2) that this influence is due to otitis, and (3) that the degree of shift to the left is a measurement of the extent of otitis influence.

No factual verification exists for these statements.

The distribution curve of my "chronic otitis" group is shown in Fig. 5. The figure shows the distribution curves of the normal material, the "chronic otitis" group and the acute otitis group in my 1940 study. It may be noted that although the "chronic otitis" curve obviously differs from the normal curve by virtue of the use of otitis as a selection factor, this deviation does not lead to the conclusion that otitis has caused the small air cell systems which may be present in a section of the group. When poverty replaces otitis as a selection factor in Tumarkin's series, the same reasoning precludes drawing conclusions that poverty has been influencing the growth or final size of the air cell systems. This observation holds true even if Tumarkin's distribution curve could be shown to be significantly skewed, which has not been done. As will later be stressed, although a shift to the left clearly exists in the curve of Tumarkin's slum group, even this shift to the left has not been shown to be definitely related to his selection factor of poverty.

Tumarkin's Contradictory Statements

In Tumarkin's critique of my investigations many contradictions may be noted. For example, concerning my normal curve distribution he says (page 150): "We must conclude that the distribution is not, and cannot be normal statistically and therefore it cannot represent 'healthy persons'." Discussing the same series he states (page 155): "It seems probable therefore that the near normality found by Dr. Diamant is ascribable to the fact that he examined a comparatively *normal* and socially homogenous population. . . ." Note the contradictory statements regarding "healthy" and "normality".

Furthermore, in the comparison of his English slum series with my series representative of the Swedish population, Tumarkin states (page 149): "the devastating effect of upper respiratory infection stands out so clearly in the A group" (i.e. in Tumarkin's slum group) "that it is at first sight difficult to understand how Dr. Diamant can have failed to detect it. In the Swedish population the explanation turns out quite simple. There was very little disease in his cases." (This last point was explicitly stated in my investigations.) Previous to this argument Tumarkin maintains (page 148) that "in every case below 6 square centimetres there are unmistakable stigmata of disease". The findings of my investigation refute this claim. In my normal curve distribution cases were included down to size zero square centimetres. My tabulations encompassed 26 ears having a size smaller that 4·20 square centimetres, and 50 ears smaller than 6·80 square centimetres whilst "stigmata of disease" were found only in a few exceptional cases. Therefore, Tumarkin's conclusion that small air cell systems invariably show "stigmata of disease" is not only refuted by my actual findings, but also by his "simple explanation" of the absence of diseased ears in my series.

My criterion for the presence of disease is the easily registered tympanic membrane perforation. Compared to this Tumarkin's choice of criteria of pathological changes in the tympanic membrane are very vague In his classification B (page 94) Tumarkin registers "Texture lack-lustre Cone of light reduced " In his classification X he registers "Drum intact but no cone of light " All these signs are recorded as manifestations of ear pathology First, these changes may not be permanent and may occur intermittently Secondly, they may be caused by factors located completely outside of the region of the ear itself, for instance by diseased adenoids, sinusitis, tubal catarrh, and so on Thirdly, the accuracy of observation of signs of this type varies with the objectivity of the examiner or examiners

Many mistakes may be noted in Tumarkin's rendition of my conclusions On page 148 and 149 Tumarkin maintains that I conclude "Thus, for example, mastoids above 15 cm ^{2} are practically immune from inflammation, where mastoids with systems below 6 cm ^{2} *although primarily normal,* are prone to inflammation with marginal perforation " In my study I do not speak of immunity or of inflammation with marginal perforation and do not use the figure 6 cm ^{2} which may be a misprint My actual statements are that central perforations are not encountered in ears with an air cell system larger than 15 cm ^{2} and that marginal perforations are not found when the air cell system exceeds 10 cm ^{2} in size

Many more examples can be cited For instance (page 147) Tumarkin maintains that I have made statements concerning normality of the mastoid air cell system My actual statement is that they are normal variants I explicitly stress that the term normal variant is a statistical term not to be used in a medical sense Tumarkin also maintains (page 138) that I conclude that "nothing that happens postpartum can alter the pattern of the mastoid structure" We may note the misleading change from "size" to "pattern of structure" We may also note that Tumarkin's quotation (page 153) indicates his knowledge of my statement "In the normal material every value may be assumed to represent a normal in the medical sense, i e not caused by disease or pathology at all This does not mean that any value *could* not have been influenced by disease That fact, however, is not given by the value itself—it must be shown and proved by other factors "

Tumarkin's discussions concerning my curves and figures are also replete with omissions in quotation For instance he has not mentioned that I myself have thoroughly discussed the slight skewness of -1 4 per cent and its significance Nor has he mentioned that the origin of his statement that my normal curve goes down below size zero corresponding to minus 2 square centimetres comes from my own computations

II. Tumarkin's Study on Poverty and Pneumatization

In his 1957 papers Tumarkin also presented a study the result of which he claimed to support his working hypothesis as well as his teleological and sociological convictions on pneumatization problems. He maintained that the small mastoid air cell system is pathological. He concluded that chronic otitis, cholesteatoma and otosclerosis are due to poverty and can be reduced in incidence by the elimination of slums.

In his study Tumarkin compared two groups of schoolchildren representative of the English slum and middle class respectively. The slum group consisted of 72 children, and the middle-class group of 99 children. The age was reported "at 13 (circa)" for both groups. The varying dimensions of the mastoid air cell system were measured with a planimeter on the lateral radiogram projection in accordance with my recommended method.

The following tabulation shows the main data of Tumarkin's study. The corresponding data of my 1940 investigations are also given.

STUDIES ON THE SIZE OF MASTOID AIR CELL SYSTEM ON THE RADIOGRAM IN LATERAL PROJECTION

Author	Tumarkin		Diamant
Selection factor	Slum school children	Middle-class schoolchildren	Patients hospitalized for scarlet fever from 1932-1938 (mandatory in Sweden).
Number of individuals investigated	72	99	178
Age	"13 (circa)"	"13 (circa)"	10-30 age group*
Sex	"Drawn from 4 dockside schools (2 for boys and 2 for girls)"	"Drawn from 2 private schools (1 for boys and 1 for girls)"	77 men 101 women
Representation of samples selected	Representative of the slum in the English population	Representative of the middle class in the English population	Representative of the Swedish population (normal material)
Mean size (square centimetres)	9·06	13·70	12·27
Range of size variation *observed* in the selected samples (square centimetres)	From 0 to about 20†	From 0 to about 29†	From 0 to 27·30
Standard size deviation (σ) *computed* for relevant group in the population (square centimetres)	4·26	5·44	5·40

*Only a comparatively small number of boys below 15 are included in this group.

† Exact figures not reported and therefore taken from Tumarkin's distribution curve (page 140).

Tumarkin's Conclusions from his own Investigation

The table clearly shows that the figures obtained in *the English middle-class group* closely conform to those obtained from my general Swedish population group, a conformity also stressed by Tumarkin In fact he explicitly confirms my computed 0 to 30 square centimetre range of size variation of the mastoid air cell system Thus, the size variation in the Swedish population seems to be comparable to that of the English middle-class population Consequently Tumarkin's statement that my normal curve distribution is "not truly normal" (page 97) must also apply to the size distribution in his middle class group

On the other hand Tumarkin claims that a significant difference exists between his slum and middle-class groups While giving the actual size distribution found in his investigation (histogram Fig 6) he maintains that "it requires no expert statistical knowledge to recognize that the two groups differ significantly as predicted by the environmental theory" The present author agrees that the two curves obviously differ, but he cannot see that they do so "as predicted by the environmental theory"

It may be recalled that the fact that distribution curves deviate from the normal curve distribution does not in itself indicate that pathological factors have been influencing growth A curve obtained from a group of children as compared with a group of adults will show a shift to the left until the growth of the air cell system is completed Curves comparing a group of boys and one of girls will show a shift up to the age of 15 A skew curve is found when the selection factor itself yields an overrepresentation of small air cell systems

The comparison of the histograms of Tumarkin's two groups discloses a marked shift to the left in the slum group However he not only maintains the existence of a shift to the left in his slum group, but also points to a skewness in the distribution curve in my distribution curve of the group representative of the general population Therefore the difference claimed to exist between his slum and middle-class group (the latter conforming to my "skewed" general population group) indicates his view, (1) that a skewness exists in his middle class group and (2) that a significantly greater skewness exists in his slum group This supposed difference in skewness between Tumarkin's two groups requires further scrutiny

The mean size of 13 7 in the middle class group may fairly well approximate the mean value of a slightly skewed normal curve distribution with the range of variation between 0 and 30 square centimetres (and a standard deviation of 5 44) as actually found by Tumarkin in this group

The mean size of 9 06 obviously cannot represent the mean of a slightly skewed normal curve distribution with the range of variation between 0 and 30 square centimetres But the reported range of variation in the slum group is between 0 and 20 square centimetres (and the standard

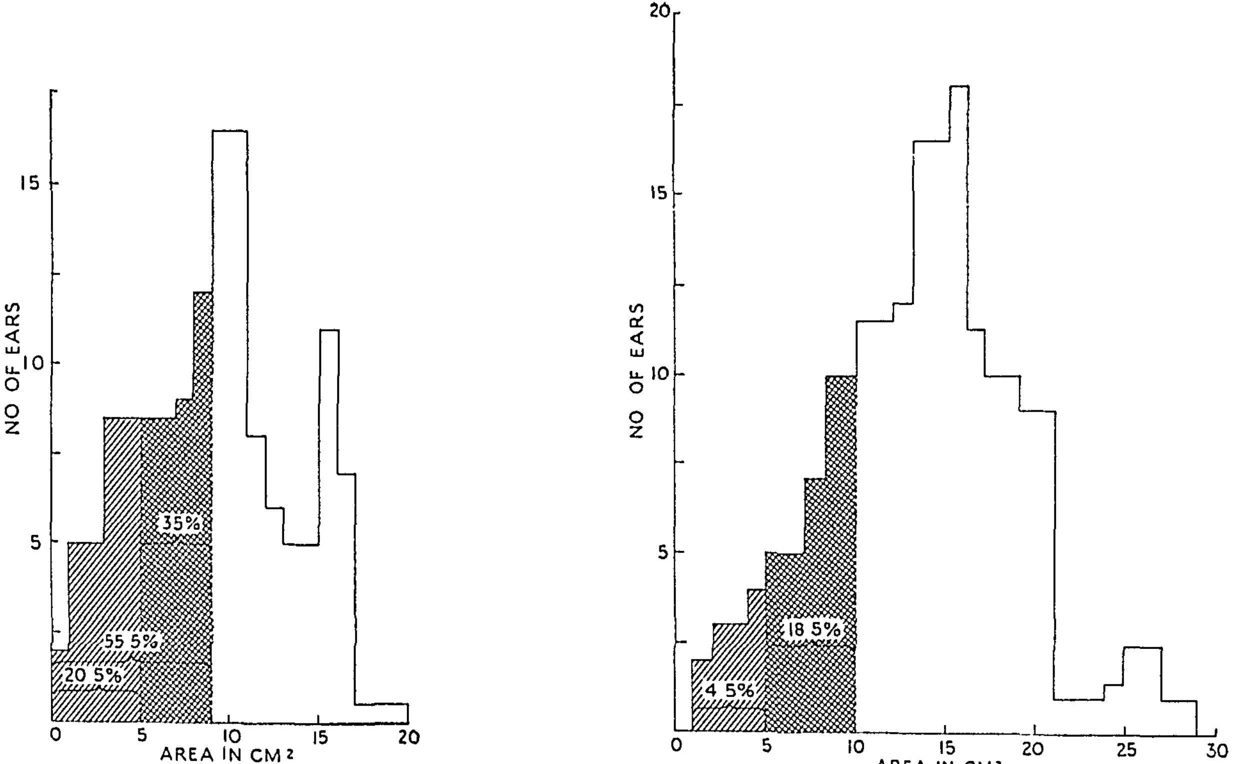


Fig 6a
(A) Distribution of Pneumatization in 144 Artisan ears at age 13 (*circa*). Mean = 9·07. σ = 4·26

Fig 6b
(B) Distribution of Pneumatization in 198 middle-class ears at age 13 (*circa*). Mean = 13·7 σ = 5·14

Fig 6

The shaded area represents mastoids up to 5 cm² which are certainly diseased The hatched area between 5 and 10 represents mastoids which may or may not be diseased Many of them are the result of aseptic inhibition Note that more than half of the (A) mastoids are probably pathological in one sense or another.

deviation is 4·26). Thus, 9·06 may fairly well approximate the mean value of a slightly skewed normal curve with this range of variation (and the reported standard deviation). It follows that the skewness in the slum group may also be fairly slight.

It may first be noted that a skewness cannot be directly observed in either of Tumarkin's two groups. This is shown by careful evaluation of his figures of mean size, range of variation, standard deviation, and of the two histograms (page 140). A difference in skewness between the two curves is even less apparent. An obvious deviation from the normal curve distribution is illustrated in the "chronic otitis" group in my graph in Fig. 5.

Tumarkin's histograms are somewhat incorrectly drawn, and do not facilitate accuracy in computations. For example, only 196 of the 198 ears are included in the histogram 11b and only 143½ of the 144 ears in the histogram 11a. The hatched area in the histogram 11a, said to cover the range of variation between 5 and 10 square centimetres, actually only covers the area between 5 and 9. The corresponding percentages are therefore incorrect. However, we may disregard these inaccuracies.

To statistically disclose a skewness in distribution curves requires a complicated analysis. The possibility of disclosing a skewness in Tumarkin's two distribution curves is obviated by many factors, one of them being the small number of samples studied.

It may be remembered that in the ideal normal curve distribution the number of samples having a size above the mean value represent 50 per cent. of the samples registered. The method derived by Lindeberg enables one to compute the magnitude of actual deviation from 50 per cent. Using this method, the skewness in my normal material (age group 10-30) was found to be —1·4 per cent.

Using the same method I have attempted to compute the deviations in Tumarkin's two groups. In the slum group (approximately) 75 samples instead of the expected number of 72 are found to be *above* the mean value. This results in a skewness of about +2·3 per cent. In the middle-class group (approximately) 100 samples instead of the expected number of 98 (two samples are not registered in the histogram) are found to be *above* the mean value. This results in a skewness of about +1·0 per cent. It is to be noted that both histograms contain more samples greater than the mean. Based on these figures the following conclusions are reached.

(1) A significant skewness in one or the other of Tumarkin's two groups has not been shown to exist.
(2) A significant difference in skewness between the two groups has not been shown to exist.

It is agreed that the *figures* arrived at in Tumarkin's slum and middle-class group differ statistically. The differences represent, however, merely a shift to the left since parallel differences are found for all three values, i.e. for mean size, range of variation and standard deviation. A difference between the mean sizes alone cannot be used to support a claim of a difference in skewness. Tumarkin, nevertheless, claims (page 223) that "the correct index is the mean" which must be a neglect of elementary statistical rules.

The question arises as to the explanation for the "shift to the left", i.e. to explain the differences in mean size, range of variation and standard deviation actually found by Tumarkin to exist between his middle-class and slum groups.

Sex and Age Distribution

The determination of significant similarities or differences between Tumarkin's slum and middle-class group is difficult, due to lack of available information in his publication regarding sex and age distribution. My investigations have shown an important sex difference in rate of growth of the air cell system, especially apparent between the ages of 10 and 15, the air cell system in girls terminating its growth at age 10 and in boys at age 15.

Therefore, if the slum group consisted preponderantly of boys and the middle-class group mainly of girls, the mean figures arrived at by Tumarkin would conform to those obtained in my investigations. In such a case any difference between the two groups would be coincidental and caused by a sampling whose composition was inadequate for comparisons.

Tumarkin's choice of age "at 13 (circa)" may also lead to differences between his two groups. In boys, it may be remembered, every single year of life up to age 15 may represent a growth of about 2 square centimetres. So, for example if the slum group consisted of children at age 12 and the middle-class group of children at age 14 we would again, due to the presence of boys in the investigation (even at a rate of 50 per cent.) have coincidental differences caused by the age sampling.

The shortening of the range of size variation from 30 square centimetres in the middle-class group to 20 square centimetres in the slum group, paralleling the decrease in mean size from 13·9 to 9·06, indicates that dissimilarities in sex sampling or age sampling or both may have caused the differences between Tumarkin's two groups.

We may, on the other hand, accept the possibility that the differences between the two groups are not coincidentally caused by sex and age incongruities. We may then discuss as to whether they may be caused by some other factor in the study; as, for instance, Tumarkin's choice of poverty as one of his selection factors.

Poverty as a Selection Factor

Ability to succeed in the community is influenced by both hereditary and environmental factors. Inherited as well as acquired disabilities are constantly influencing the composition of the slum and middle class in the population. Disabilities are keeping individuals in the slum and are constantly adding individuals to the slum. Slum and middle class therefore will differ in number of disabled persons. Disabled persons are reproducing and their children will consequently be born and go to school in the slum.

There are many causes for disability, one of the causes being various kinds of disease. Hard of hearing is an infirmity related to ear disease. Chronic middle-ear discharge occurs more frequently in individuals having small air cell systems. As previously mentioned, tympanic membrane perforations do not occur in ears having an air cell system exceeding the mean size computed for the general population.

Dahlberg and I have shown that the final size of the mastoid air cell system is determined preponderantly by hereditary factors. Tumarkin disagrees with this conclusion, supporting his reasoning by an arbitrarily and mathematically incorrect interchange of figures obtained in different samplings of my investigations. He, however, still admits a "residual genetic variation" (page 149) which he claims will become operative in a "Utopian State". Thus the importance of heredity in determination of the final size of the mastoid air cell system is in fact accepted by all investigators, Tumarkin (and Wittmaack) included. Consequently, since hereditary factors are operating to some degree, an extra number of individuals with small air cell systems may be brought into the slum group by Tumarkin's choice of poverty itself as a selection factor. Therefore, the slum group may show a larger number of small air cell systems than the middle-class group. This may explain why Tumarkin's slum group includes sizes down to zero, whilst the middle-class group does not. Furthermore, since the presence of small air cell systems is often associated with occurrence of discharging ear diseases and changes in the tympanic membrane, hearing deficits will also be found more frequently in the slum group.

Tumarkin finds that his selection factor exerts a negligible effect. He states (page 141): "It is difficult to believe that there can be any significant genetic difference between two such adjacent social strata, especially when that stratification has only taken place within the last hundred years". This statement seems to constitute weak support for a denial of hereditary influences, especially since the importance of hereditary factors in the growth of the mastoid air cell system has been accepted in all investigations on pneumatization problems, and, moreover, is admitted by Tumarkin himself.

Hachfeld (1954) argued that poverty causes a proneness of poor people to contract infantile infectious otitis, and thereby effects a stunting of growth of the mastoid air cell system. In a discussion of this special relationship, I stressed (Diamant, 1955) the fact that no extant investigations had shown any influence of poverty on mastoid air cell system growth. I also stressed that even if future investigations could show that poverty really exerts such an influence, it would still remain to be shown that this influence takes place by way of otitis. No studies on this problem have appeared.

In my studies using the presence of tympanic membrane perforations as a selection factor, a markedly deviated distribution curve was obtained and no conclusions could be drawn regarding factors of growth of the mastoid air cell system. Tumarkin's study, with poverty as one of the selection factors, shows no skewness or difference in skewness in his two groups. Even if these differences did exist, they could not substantiate his conclusions on the causative factors of growth. Differences shown to exist in tympanic membrane changes and in hearing cannot support such conclusions. Finally, even if poverty can be shown to exert an influence on development and growth of the mastoid air cell system, such an influence cannot be equated to an influence of otitis or upper respiratory infections.

It may be added that my conclusion that the final size of the mastoid air cell system is decided by hereditary factors and prenatal environmental factors cannot be invalidated by Tumarkin's argumentation (page 98): "This is a remarkable conception especially when we remember that before birth the air cell system does not exist." The size and shape of the teeth, for instance, being preponderantly inherited, must have been decided prenatally in spite of the fact that the growth takes place after birth. The same holds true of the post-natal growth of the nose and other characteristics of final size and shape, all being genetically influenced. There is nothing remarkable in the fact that growth continues after birth. Tumarkin's statement seems to indicate that growth after birth must be caused by environmental factors exclusively, not to say by pathological environmental factors.

Furthermore, it is quite obvious that Tumarkin's statement (page 98) that I have not determined the exact nature of the causative environmental factors, is extraneous to the discussion. Any such determination lacking support from scientific investigations would of course be mere guesswork. Tumarkin's proclamation that they are pathological and represent otitis must be presumed to be founded on his investigation on poverty and pneumatization.

Pathology and Size of the Mastoid Air Cell System

Tumarkin indicates that the small sizes of air cell system found in the general population are pathological, and induced in the following ways:

(1) They may be inhibited before development.
(2) They may be halted at any time during development.
(3) They may be diminished by varying degrees of destruction during development and after completed growth.

It has already been stressed that none of these possible influences has been shown to have caused the shift to the left actually found in the slum group as compared with the middle class group Assuming that a pathological influence has actually been present, a shift to the left may be expected to be obtained when all individuals are influenced, and to about the same extent The pathological shift to the left would then imply that all sizes of air cell system (possibly except only the largest one) are pathological It is true that the shift to the left actually found by Tumarkin in the slum group would be the result of such a pathological influence on growth But this seems not to conform with his three different ways assigned for pathology to influence growth Neither does it conform with Tumarkin's claim (on page 157) "In any case however, as we have already seen, it is impossible to regard mastoid asymmetry as a biologically normal condition " Asymmetry can hardly represent the effect of pathological factors influencing all sizes to about the same extent

In claiming that asymmetry should be judged according to percentages of the actual size, Tumarkin (page 216) argues that asymmetry of 4 cm² in large air cell systems is "normal and symmetrical' His choice of examples for supporting this view however, disregards the fact that an asymmetry of up to 8 square centimetres may occur in individuals having the smaller air cell system as large as 15 square centimetres (thus having both air cell systems above the mean size of the general population and above the lower border claimed by Tumarkin for the 'cellular i e the 'normal' mastoid) It may in this connection be mentioned also that in my investigation on samples representative of the general population the rate of asymmetry, registered in square centimetres, forms a continuous sequence and shows a normal curve distribution This is an additional strong support for the "anatomical normal variant theory"

It is essential to determine what size Tumarkin designates as a small air cell system Tumarkin in his article states

(1) That sizes below 4 square centimetres are pathological (page 142)

(2) That sizes below 5 square centimetres are certainly abnormal and up to 9 or 10 are probably so (page 161)

(3) That sizes below 6 square centimetres are pathological (page 240)

(4) That 6 square centimetres is the lower limit for a 'normal' mastoid (page 160)

(5) That sizes below 15 square centimetres are hypocellular, above 15 square centimetres are cellular and above 20 square centimetres are hypercellular (page 96)

(6) That any size may be pathologically halted by otitis processes before completed growth (page 245)

(7) That any size may be pathologically diminished by otitis processes after completed growth (page 98).

Add to this that Tumarkin claims:

(8) That an anatomical size variation must be accepted (pages 137 and 221).

(9) That the size is influenced genetically (page 132).

According to the theory of "halted growth", an air cell system of say 25 square centimetres may have been halted at that size. According to the "secondary obliteration" theory it may also be the result of a reduction from a size of 30 square centimetres. Logically, this final size may also be anatomically determined without the influence of pathological factors. The same holds true for any size along the whole range of variation. Consequently, the final size is not itself decisive and no size can be excluded from being pathologically influenced.

An inconsistency is noted in Tumarkin's attempt to designate a border between pathological and non-pathological sizes. It is illogical to presume that a halting of growth may occur during the whole developmental growth period up to completed growth at age 14 (page 245) and that a secondary obliteration of growth may occur at any time after birth (page 98), while at the same time designating a dividing line between pathological and non-pathological sizes. Tumarkin's own views should, if correct, prohibit him from setting up such a border. It is still more illogical to designate so many varying borders, not to say choosing them so arbitrarily.

The sizes between 15 and 20 square centimetres must furthermore be presumed to be "normal" according to the nomenclature chosen by Tumarkin. Tumarkin writes (pages 244): "The cellular mastoid is primarily normal, i.e. 'healthy'." Thus "normal" mastoids are those "cellular" ones comprising sizes between 15 and 20 square centimetres. "Hypocellular" explicitly means pathological. It is stated (page 87) that "hypercellular" means "normal". In any case, it cannot be a question of a pathological increase of size, since Tumarkin claims that pathology means inhibition or secondary obliteration (page 98). (Besides, the otological literature claims that hypercellular air cell systems may occur in cases of acromegalia.)

Tumarkin proclaims the existence of a group of Wittmaackians, and lists himself as a member of the Neo Wittmaackians. Wittmaack, however, does not belong to any of these groups. Wittmaack himself denies explicitly that his conclusions concern the *size* of the air cell system. Instead they are claimed to concern *irregularity*. This irregularity according to Wittmaack may occur in *all* sizes of air cell system, from the very smallest to the very largest. Wittmaack's investigations, therefore, cannot be used to support theories concerning growth and *size* of mastoid air cell system in the general population.

Tumarkin also claims that the investigation recently published by

Friedmann (1956) supports the theory of a pathological influence on the final size of mastoid air cell systems Friedmann's experimental work, however, was concerned with destruction of the bulla in guinea pigs and does not support Tumarkin's statement concerning the halting of growth at any time during development

Many additional reasons may be given to show why Friedmann's studies on the destruction of the bulla in guinea pigs cannot be used to support the theory of a pathological influence in final size determination in man It is true that it cannot be excluded theoretically that otitis in man may cause a local destruction of single air cells, or air cell groups, or even of a total air cell system Such destructions must, however, be supposed to have happened unobserved in man if they, ex analogiam from Friedmann's findings, should be maintained to be valid for the development of small air cell systems in the general population I say unobserved, because they have not been observed in manifest cases of air cell destruction, not even in acute mastoiditis resulting in mastoidectomy, often seen in the recent past On the other hand, even if such destructions could occur clinically unobserved, which as stated is theoretically possible, they must in any case always lead to additional clinical signs of the disease process, otoscopically, functionally and radiologically Such signs are almost constantly missing in the small mastoid air cell systems of the general population Consequently the conclusion that mastoid air cell destructions clinically showing no signs whatsoever of ear disease, represent actual occurrences in the small air cell systems of the general population, showing no otoscopic, functional or radiological signs of residual ear pathology in a scrutinizing investigation cannot rest on scientific grounds

I would like to quote another of Tumarkin's unsubstantiated statements (page 157) "Now let us examine a hypocellular mastoid—say of 2 cm ^{2} It is obvious that the intercellular trabeculae are much thicker than the trabeculae of the hypercellular mastoid and in the writer's opinion, this alone justifies us in labelling the former 'abnormal' " As a matter of fact the trabeculae are not thicker in the smaller air cell systems, at least not in the normal material in my investigation Even if they were thicker in Tumarkin's case material (which he has not shown to be the case), this still would not justify conclusions concerning their "abnormality" If thicker trabeculae are to be proven pathologically caused, they must show the microscopical stigmata of actual or previous inflammation

Furthermore, in all scientific investigations definitions must meet rigorous standards Statements claimed as basic premises by Tumarkin, such as ' the normal mastoid is always fully pneumatized" have no scientific meaning The word normal cannot be used in a medical sense, and "fully pneumatized" gives no idea of the size of the air cell system In my 1952 monograph, *Chronic Otitis*, I discussed (Diamant, 1952) these ambiguous expressions, space does not permit a further refutation. May I only add that Tumarkin states (page 153) my deduction that "completely acellular mastoids are to be regarded as normal". What I actually say is that they may be regarded as normal variants. Consequently the whole argumentation following this false quotation is based on false premises.

Suggestive nomenclature should also be avoided in the scientific literature. Tumarkin's term "not truly normal" has already been discussed. It gives the impression that an absolute conformity with a normal curve distribution can be reached. The nomination of "hypocellular" air cell systems to represent small size systems suggests that the development of small air cell systems has been determined by pathological factors. Use of this term prejudices the argument, i.e. it is impossible to prove that hypocellular (small) air cell systems are pathological by stating that they are small. However, this is what Tumarkin does. Moreover, the use of the term "hypocellular" for small air cell systems should automatically lead to a use of "hypercellular" when representing large air cell systems. Large air cell systems are not designated as pathological by Tumarkin, but are claimed to be healthy (page 87). Above all, arbitrary nominations of borders between "pathological" and "normal" sizes of air cell system offend laws of scientific reliability.

A few words remain to be said about Tumarkin's teleological and sociological convictions, since they seem to play such a great part in his effort to support his conclusions.

According to Tumarkin's teleological convictions, any anatomical formation, as for instance the mastoid air cell system:

(*a*) Must have a function.
(*b*) This function must be known.
(*c*) Agenesis, having no function, it cannot be a hereditarily or anatomically caused variation, but must result from pathological influence.

The following quotation from Tumarkin's study (on page 156) seems to be very elucidating: "Take for example, what must surely be a dominant criterion in the conception of normality—function. It goes without saying that a normal organ must possess a normal function. Now the precise function of the pneumatic system is not clearly known, but enough has been said to suggest that perfect functioning of the mammalian middle ear is impossible in the absence of accessory air spaces. If that is accepted it follows automatically that the acellular mastoid lacks its correct function and therefore it must be abnormal."

As illustrative of his teleological reasoning, Tumarkin argues that the small air cell system has been shown to be pathological by proving that it is actually small (page 157).

I would, first, like to mention that in my series of samples representative of the general population, there are included individuals with ears having no mastoid air cells at all, and nevertheless showing no signs of ear pathology whatsoever.

Second Tumarkin's equation of agenesis and pathology is also refuted by the following findings by Longo and Blandino (1956).

"The authors by means of a genealogical study inquire as to the importance of hereditary factors in the mechanism of pneumatization of sinusal cavities. They examined 42 subjects of 2 different families and extend the investigation to 3 generations. In the investigation of these 42 subjects the authors found the agenesis of frontal sinuses occurring in 70 per cent. and of sphenoidal sinuses in 59 per cent. The occurrence of the agenesis has been found in 5 per cent. and 2 per cent. respectively in the general population. The results demonstrate the hereditary origin of agenesis. The transmission modality of agenesis seems to be multifactorial according to the author's opinion."

As for Tumarkin's sociological convictions, he seems to claim that any disease occurring more commonly in poor than in rich people signifies:

(*a*) That it is caused by poverty (consequently disease never causes poverty).

(*b*) That poverty as a causative factor means otitis infections and upper respiratory infections.

(*c*) That heredity can be excluded as a causative factor.

It would require too much space to fully discuss conclusions supported by sociological and teleological reasoning. May I only mention Tumarkin's comment that my conclusions concerning causative factors of size variation of the mastoid air cell system will be true in a "Utopian State" created by the elimination of the slums.

It may be stressed, finally, that Tumarkin's suggestion that *otosclerosis* is caused by poverty and otitis infections is based on the very findings in his 1957 study, and follows the same type of reasoning pathways.

Summary

Tumarkin in his investigation in 1957 concerning pneumatization problems claims to have shown:

(1) That poverty determines growth (and agenesis) of the mastoid air cell system.

(2) That poverty in this connection is equivalent to otitis infections and upper respiratory infections.

(3) That poverty causes otosclerosis.

In my opinion scientific investigations on pneumatization problems must provide for:

(1) Adequate selection factors for the group sampling.

(2) Adequate non-suggestive nomenclature.

(3) Adequate scientific reasoning and conclusions not reaching far beyond the limits of the investigation.

Tumarkin's investigation, even were it not offending these three scientific premises, cannot reach valid conclusions concerning *causative* factors in the size variation of the mastoid air cell system.

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HISTOCHEMISTRY OF THE NASAL RESPIRATORY MUCOSA

By MILES TAYLOR (Sheffield)

Introduction

SCHNEIDER (1660) postulated that the nasal secretions are a product of the membrane lining the nasal airways, and Ingelstedt and Ivstam (1949) showed that they are purely glandular in origin. The morphology of these glandular structures has been well described, notably by Zuckerkandl (1882), Von Heiss (1931), Schaeffer (1932), Brunner (1942) and Leonardelli, Pizzetti and Chierego (1952), and their normal variations by Proetz (1941) and Hollander (1944).

The chemical composition of the nasal secretions was first studied by Russell and Gies (1906), using physical methods on collected secretions. Schaeffer (1932) and later Buhrmeister (1936) made observations on the mucin of the upper respiratory tract. Lillie (1949) mentions the histochemical characteristics of the mucopolysaccharides in the basement membrane of the upper respiratory tract, and de Vanna and Salonna (1953) described PAS-positive granules in the glands of the nose, but no systematic histochemical studies have been made on the nasal respiratory mucosa.

In this paper the results of a comparative study in man, rabbit and rat are presented as a preliminary to the investigations of pathological nasal states.

Materials and Methods

Human nasal mucosa was obtained from the septa and turbinates of patients undergoing operation under general anæsthesia, nasal respiratory mucosa was also obtained from the septa of freshly-killed adult male and female rabbits and rats. Each specimen was divided into two, one part being fixed in ice cold acetone and the other ice cold 80 per cent. alcohol. Paraffin sections were made, and apart from routine staining with hæmatoxylin and eosin, the following stains were used: iron hæmatoxylin and picrofuschsin (Van Gieson) and Masson's trichrome for the demonstration of collagen, Gordon and Sweets' technique for reticulum, Southgate's mucicarmine for mucins, Alcian blue for acid mucopolysaccharides and the periodic acid-Schiff (PAS) technique as modified by Hotchkiss (1948) for the demonstration of mucopolysaccharides.

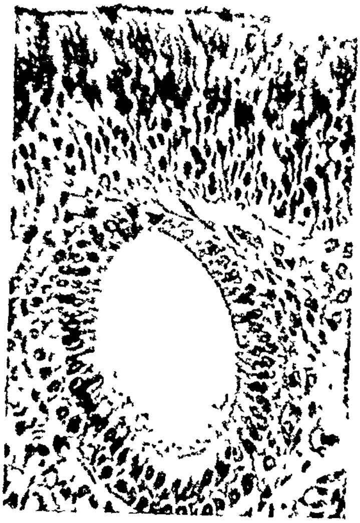


FIG 1

Man Pseudo-stratified columnar ciliated epithelium, goblet cells, basement membrane "basal canals", and large duct PAS counterstained with hæmatoxylin × 150

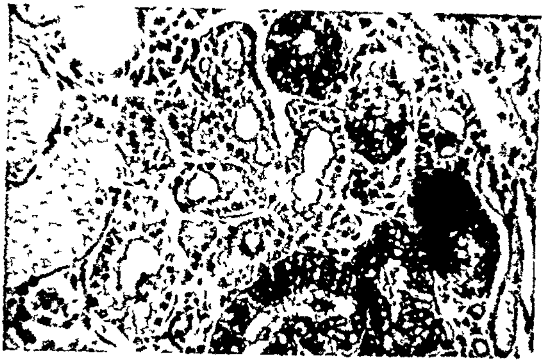


FIG 2

Man Serous and mucous glands PAS counterstained with hæmatoxylin × 100

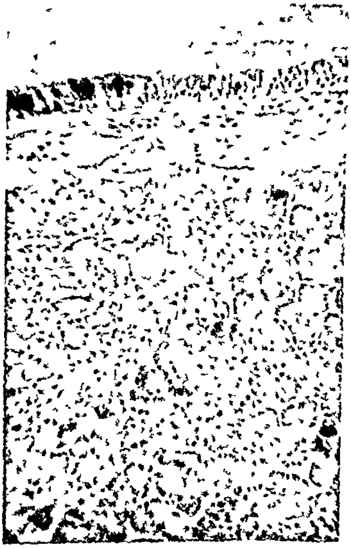


FIG 3

Rabbit Anterior mucosa Ciliated columnar epithelium, goblet cells and two types of gland PAS counterstained with hæmatoxylin × 50

FIG. 4.

Rabbit Posterior mucosa Columnar ciliated epithelium and simple tubular glands PAS counterstained with hæmatoxylin × 80

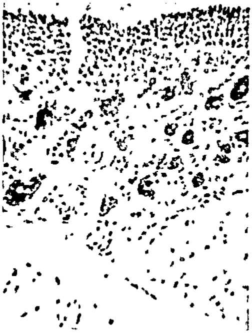


FIG 5.

Rabbit Anterior mucosa Goblet cells and glands *Alcian blue counterstained with Safranin. ×200*

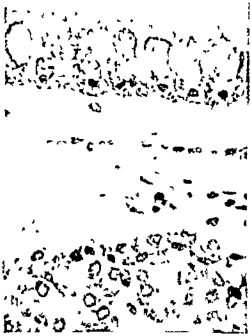
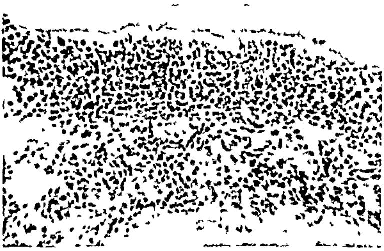


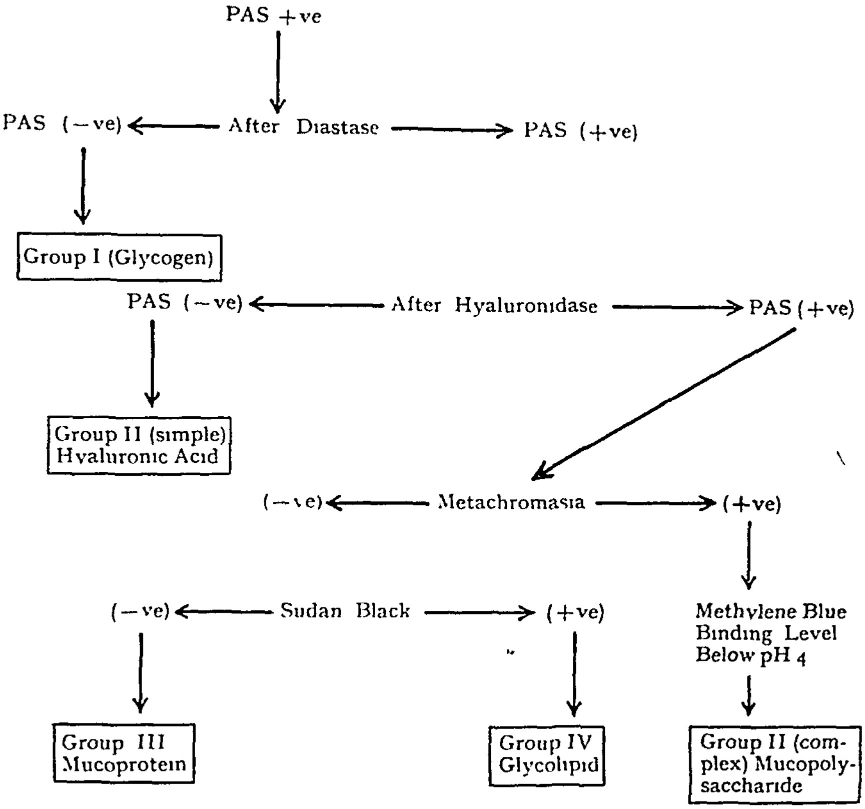
FIG. 6.

Rabbit Posterior mucosa Simple tubular glands Alcian blue counterstained with Safranin × 80



The PAS-positive material was differentiated according to the following scheme, based on G. B. Roberts and W. F. H. Jarrett (personal communication to Professor F. Davies):

THE IDENTIFICATION OF PAS POSITIVE MATERIAL



The Feulgen reaction (Pearse, 1953) and the methyl green-pyronin technique (Kurnick, 1955) were used to identify deoxyribonucleic acid (DNA), and ribonucleic acid (RNA) respectively. Non-specific esterase (Pearse, 1953) was demonstrated by the α-naphthol acetate method. Alkaline phosphatase was demonstrated by the calcium-cobalt method of Gomori (1946), with the addition of magnesium ions to activate the enzyme as suggested by Kabat and Furth (1941). The substrate used was sodium β-glycerophosphate and the incubation period one hour.

Cholinesterase was demonstrated by the myristoyl choline method (Pearse, 1953).

RESULTS

The thickness of the nasal mucosa varies with its position in the nasal cavities. It is thinner, less vascular and more uniform in the rabbit and rat than in man, in whom the variation in thickness is marked.

The nasal respiratory epithelium in man (Figs. 1 and 7) is composed of tall columnar ciliated cells with nuclei in several planes, giving a pseudostratified appearance. The cytoplasm of some of the cells is filled with mucus. Aggregations of mucus-secreting cells are sometimes seen and these constitute the intra-epithelial glands. The epithelial cells lie on a well-defined basement membrane, which varies in thickness in different places and is pierced by "basal canals". The latter have a thin endothelial lining and show the typical signet-ring appearance of blood capillaries. They give a reaction of variable intensity for alkaline-phosphatase, and serial sections show them to be part of the sub-endothelial capillary plexus.

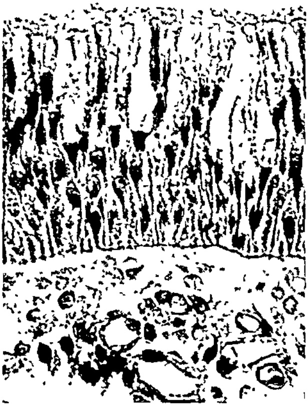


FIG. 7

Man columnar pseudo-stratified ciliated epithelium, goblet cells, basement membrane and "basal canals". Hæmatoxylin and eosin. × 430.

The most striking feature about the epithelium on the *anterior* part of the septum of the rabbit and rat is the presence of a well-marked layer of basal cubical cells with large, round, central nuclei (Figs. 8 and 9); this layer is more marked in the rabbit than in the rat. The columnar ciliated cells are not so tall as in man, and the cytoplasm of some of the cells is distended with mucus. Unlike the human, there is no basement membrane.

The glands in the sub-endothelial tissue are compound alveolar in type in man, rabbit and rat. In man (Fig. 2) two types are seen, serous and mucous; both may be present in one alveolus. The component cells of the

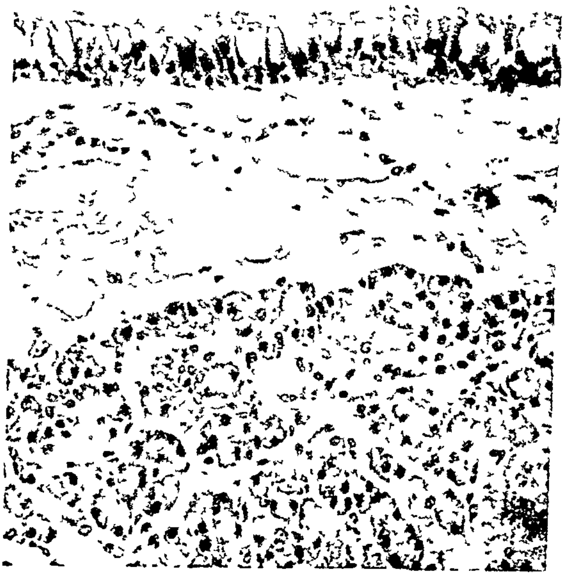


FIG 8

Rat Anterior mucosa. Columnar ciliated epithelium, goblet cells, basal layer of cubical cells and glands Hæmatoxylin and eosin × 250

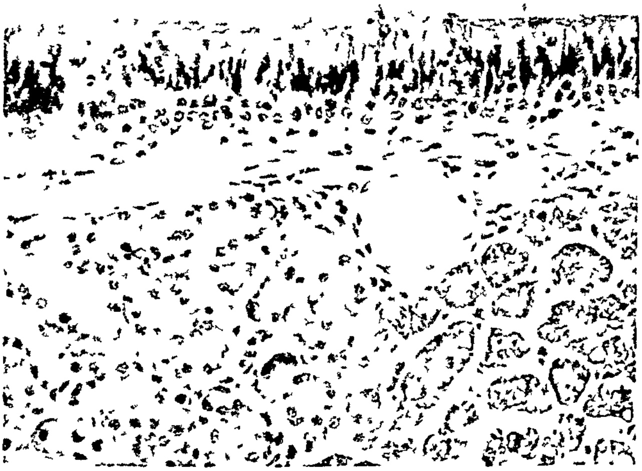


FIG 9

Rabbit: Anterior mucosa Columnar ciliated epithelium, basal layer of cubical cells and two types of glands (see text) Hæmatoxylin and eosin × 250 serous glands have large, round nuclei lying towards the bases of the cells, the cytoplasm of which is filled with fine granular material. The cytoplasm of the mucous glands is distended with mucus and is vacuolated· the nuclei are flattened against the bases of the cells.

In the rabbit (Fig. 3) and rat, the glands approximate to the serous type of gland seen in man. The cells are regular in outline with large round nuclei lying towards the bases of the cells, and the cytoplasm is filled with fine granular material. The large ducts are composed of cubical cells with round, centrally placed nuclei. The cytoplasm of the cells is less eosinophilic in man than in the rabbit and rat.

In the rabbit and rat, the nasal respiratory mucosa on the posterior part of the nasal septum shows considerable difference in morphology from that described above for the anterior part. The epithelium is ciliated but is taller and the nuclei are in several planes, giving a pseudo-stratified appearance (Fig. 10). The basal, cubical layer of cells is not well defined and in places appears to be absent. The glands are simple tubular structures, the component cells of which are irregular in outline, with round, centrally placed nuclei. They open on to the surface of the mucosa by a duct lined with a single layer of cubical epithelium.

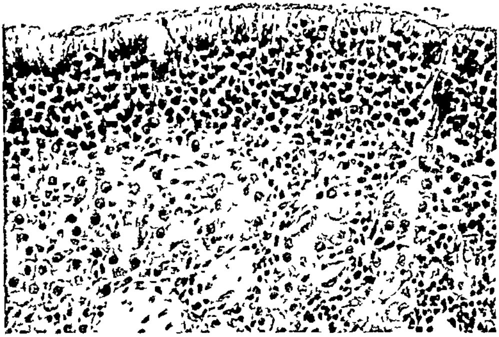


FIG 10

Rabbit Posterior mucosa Pseudostratified columnar ciliated epithelium Simple tubular glands and ducts Hæmatoxylin and eosin × 250

Histochemistry

In man, the cytoplasm of some of the epithelial cells gave a strongly PAS-positive reaction. The basement membrane was PAS-negative The mucous glands were also strongly positive, while only the superficial part

The results obtained for cholinesterase were inconsistent in all three species.

In man alkaline phosphatase was found only in the blood vessels. In the rabbit a positive reaction was seen in the cytoplasm of most of the basal cubical cells, but in the rat phosphatase was present in the cytoplasm of all layers of the epithelium.

Discussion

Although Swindle (1935) and Petrillo (1950) described a sub-endothelial capillary plexus in the nasal mucosa of mammals, other authors considered that in man the "basal canals" are fenestra in the basement membrane. However, serial sections of the human material in the present study, confirm that the "basal canals" are part of the sub-endothelial capillary plexus, with the typical signet-ring appearance of capillaries that, as in many other parts of the body, give a positive reaction for alkaline phosphatase.

The glandular elements of the nasal respiratory mucosa are shown to produce a secretion containing both muco-protein and acid mucopolysaccharide. This supports the work of Russell and Gies (1906), but these authors were unable to determine whether the material was mucoprotein or nucleo-protein.

It is generally considered that basement membranes are composed of an amorphous PAS-positive material associated with reticular fibres (Ham, 1957). Lillie (1949) found the basement membrane of the upper respiratory tract in man to be PAS-positive. In the present work repeated experiments under carefully controlled conditions have not confirmed Lillie's findings for the nasal respiratory mucosa, but a dense network of reticular fibres was seen in the basement membrane after using Gordon and Sweets' technique for reticulum.

The presence of alkaline phosphatase in the respiratory epithelium of the rabbit and rat is an unexpected finding. In epithelia such as those in the intestine and kidney, the presence of phosphatase is usually correlated with absorption. Although in other situations, such as ossification centres, phosphatase is found where the transport of organic molecules is not apparently involved, no reason can be suggested for its existence in sites demonstrated here.

Reynolds and Foster (1940), using physiological methods, were unable to demonstrate the presence of acetylcholine in the nasal respiratory mucosa of the rabbit. The present histochemical studies confirm this finding.

Summary

The normal histology of the nasal respiratory mucosa of man, rabbit and rat is described. In man, the cytoplasm of the epithelium of the entire

nasal respiratory mucosa contains PAS-positive material; in the rabbit and rat, however, this material is limited to the anterior part of the mucosa. All the mucosal glands in man contain PAS-positive material and stain heavily with mucicarmine. While the glands in the posterior mucosa in the rabbit and rat contain PAS-positive material and stain with mucicarmine, those in the anterior mucosa, though PAS-positive are mucicarmine negative. The basement membrane in man is PAS-negative but contains reticulum. No basement membrane was found in the rabbit and rat.

The present work has shown that the PAS-positive material in all the above sites contains mucoprotein and mucopolysaccharide. A strong reaction for non-specific esterase is noted in the anterior epithelium and simple tubular glands of the rabbit and rat. The remaining glands, like the posterior epithelium, show a weaker reaction, while in all these tissues in man the reaction is negative.

Acknowledgments

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PASSAGE OF FLUID AND OTHER SUBSTANCES THROUGH THE NASAL MUCOSA

By J M YOFFEY* (Bristol)

On the last occasion when I had the privilege of addressing a similar gathering of otorhinolaryngologists, I was engaged in the preparation of a film on the lymphatic system, part of which dealt with the lymphatic drainage of the nasopharynx The film was then unfinished, and I thought, therefore, it might be appropriate on this occasion if I prefaced my remarks with the completed film—or at any rate, this particular section of it

I do so not merely to show the film as such, but to emphasize one point which the film happens to bring out rather clearly In the experiment in which we had cannulated the deep cervical duct in the monkey, the cannula was placed vertically You may have noticed that after the nasal instillation of a solution of Evans Blue, the solution on reaching the cannula at once rose to the top The reason for this, not mentioned in the commentary, was that the dye had been dissolved in distilled water, and the speed of absorption must have been such that though no doubt some salts would have been taken up by the distilled water in the tissue spaces and the lymph, there had not been enough to make the solution isotonic Being therefore, still hypotonic, it would be lighter than the previously formed lymph and so would float to the top Such at any rate would appear to be the most likely explanation, and it serves to emphasize the fact that the nasal mucous membrane seems to permit, with great readiness, the passage of distilled water

This may on first consideration appear to be a somewhat singular phenomenon, more especially since normal saline is almost completely unabsorbed after nasal instillation The basic facts concerning the absorption of water and saline were first demonstrated clearly by McCarrell (1939) Lymph from the nasopharynx was collected by cannulae placed in the right and left cervical lymph ducts McCarrell found that if the nasopharynx was irrigated with distilled water, there was an immediate and significant increase in the volume of cervical lymph which could be collected This in itself, of course, need not necessarily indicate water absorption, but at the same time the protein concentration of the lymph became more dilute. These two findings together seem definitely to point to the absorption of water. In this, and similar experiments, there is also evidence to show that some of the fluid which traverses the nasal mucosa may be absorbed directly into the blood stream, through the walls of the submucous blood capillaries.

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A number of other crystalloids can be shown to be absorbed in small amounts. Thus Blumgart (1924), under experimental conditions in which the nasopharynx could be the only possible site of absorption, found that after nasal instillation potassium iodide and phenolsulphonphthalein could be detected in the urine, while the absorption of pituitary extract could readily be shown by its immediate physiological effect (Blumgart, 1922).

Somewhat larger molecules such as the vital dyes, Trypan blue and Evans blue, could also be shown to pass through the nasal mucosa in experimental animals; and in fact they afforded an excellent method of demonstrating a living lymphatic pathway, functioning under normal conditions of flow and pressure (Yoffey and Drinker, 1938).

Still larger molecules, such as egg albumen, are also absorbed in amounts which, though small, can be readily detected by immunological methods (Yoffey, Sullivan and Drinker, 1938); similarly with serum albumen. But the larger molecules of horse serum do not traverse the mucosa, even if a sterile inflammation has been produced. Dserzgowsky (1910) found immunity to diphtheria could be produced in horses by nasal sprays and plugs soaked with toxin: Blumenau (1911) obtained a similar result. Peters and Allison (1929) induced immunity to scarlet fever by nasal spray, though some inhalation and swallowing also occurred.

The question at once arises to what extent the absorption of small amounts of protein may be responsible for such clinical conditions as hay fever. It has, in fact, been noted that in animals sensitized to horse serum, nasal instillation of horse serum can cause sneezing and discomfort. The local nature of the reaction, due to the absorption of only small amounts, can readily be shown in such cases by the fact that fatal shock follows the intraperitoneal injection of adequate amounts of serum.

Viruses present a particularly interesting problem. Virus particles do not immediately pass through the nasal mucosa. But if the animal is susceptible, they first establish themselves in it and multiply. Subsequently they enter the submucous lymphatics and pass down the cervical lymphatic pathway, where they are first held up in a lymph node. But since the virus particles are cytotropic they enter the cells of the lymph node, mainly lymphocytes, and as these latter become free and pass into the efferent lymph, they are carried by lymphocytes into the blood stream and so become rapidly disseminated throughout the body. This sequence of events was first demonstrated for vaccinia virus in the rabbit by Yoffey and Sullivan (1939). It probably holds good for other virus diseases such as the common cold Yoffey and Drinker (1939b) tried to demonstrate the same course for the virus of poliomyelitis, but were unable to do so However, work done in recent years certainly seems to make such a mode of spread rather more probable (for fuller discussion see Yoffey and Courtice, 1956)

Blood Supply in Relation to Mucosal Function

The blood supply of the nasal mucous membrane appears to be designed to discharge two distinct functions For the purpose of warming the inspired air, a copious flow of blood is required but this need not necessarily pass through a capillary network For this purpose the arteriovenous anastomosis is eminently suitable, and Harper (1949) has pointed out that there are many of these anastomoses, particularly under the mucosa of the inferior turbinate bone This is what one might expect, for despite statements to the contrary, it appears highly probable that in the adult the main air current on normal inspiration is through the lower part of the nose

But the arteriovenous anastomosis, while admirable from the point of view of heat loss would not permit the passage of fluid, which must occur freely and rapidly if the inspired air is to be saturated with water vapour Hence the so called respiratory part of the nasal mucosa must not only possess abundant arteriovenous anastomoses, but also a rich capillary network By contrast, the olfactory portion does not require the arteriovenous anastomoses of the respiratory area, and being out of the main inspiratory air current it does not need a rich capillary supply for the purpose of moistening the inspired air However, it appears to be important that the free olfactory receptors should be kept moist, and this is further ensured by Bowman's glands on the one hand, and the cerebrospinal fluid on the other

It is tempting to speculate that it is the need for moistening the inspired air which has resulted in the respiratory mucosa being freely permeable to water—but not, for obvious reasons, to the blood crystalloids As has already been pointed out, however, some crystalloids are absorbed, even if only in small amounts, and once through the mucous membrane they pass readily into both blood and lymph capillaries As is usual with such substances the bulk of the absorbed material goes into the blood stream

Changes in the Fluid Balance of the Submucous Tissues

The mucous membrane of the nose is exposed to a wide variety of insults, physical, chemical, biological It receives the first impact of the outside air at widely varying temperatures It is the primary target of irritant substances which may be inhaled, and also of living particles which can then multiply in it. The blood capillaries, therefore, can show rapid and marked changes in permeability. A sudden increase in this permeability give rise to a submucous œdema which is felt subjectively as a sensation of tightness and of blockage. The close connection between the mucous membrane and the underlying rigid bone ensures rapid rise in tissue pressure when an excess of tissue fluid is formed. This rise in pressure promotes the flow of lymph and so helps to drain away excess protein-containing extravascular fluid from the nasal mucosa.

This type of change was demonstrated very convincingly in the experiments of Cameron, Courtice and Short (1947) who applied the arsenical vesicant Lewisite, to the nasal mucous membrane and then observed changes in lymph flow. "The resulting œdema greatly increased tissue tension, so that the lymph flow often became spontaneous. . . . The protein content of the lymph rose until it reached the level in the plasma." This type of change—increased capillary permeability ————> submucous œdema ————> increased tissue tension ————> augmented lymph flow relieving the tension—would almost appear to be a self-regulating mechanism, and constitutes a sequence of events which is found after infection or other irritation. Under all these circumstances the *flow of lymph along the cervical pathway may be promoted not only by* the increased tension, but also by the pulsation of the dilated blood vessels, which may provide a pumping mechanism (cf. Robison, 1950).

Communication with the Subarachnoid Space

The precise nature of the communication between the subarachnoid space, and the submucous tissue spaces of the nose has long been the subject of controversy and experiment. Weed (1914) came to the conclusion that the absorption of cerebrospinal fluid was "a twofold process, being chiefly a rapid drainage into the great dural sinuses, and in small part a slow indirect escape into the true lymphatic vessels". One of the chief regions for this indirect escape was the olfactory part of the nose. Key and Retzius (1876) first showed that substances injected into the subarachnoid space escaped into the lymphatics of the nasal mucosa and thence into the cervical lymphatics, and this observation has since been confirmed by many workers (see for example the review by Weed, 1914; Yoffey and Drinker, 1939a). We do not know how much of the cerebrospinal fluid drains along this route, but it would appear to be a valuable additional safeguard for moistening the delicate olfactory mucous membrane.

Courtice and Simmonds (1951) endeavoured to obtain some idea of the extent to which a naturally occurring contaminant of the cerebrospinal fluid, such as plasma protein or blood, escaped *viâ* the cervical lymphatic pathway. Into the cisterna magna of the cat they introduced dye-labelled plasma protein, reaching the same order of concentration in the cerebro spinal fluid as might occur in patients with meningitis After 4-5 hours they compared the amount absorbed into blood, cervical duct lymph, and thoracic duct lymph, and found 5 per cent in the cervical duct lymph, 15 per cent in the blood, and 0 4 per cent in the thoracic duct lymph Though the greater part of the dye had entered the bloodstream by a route other than the lymph, it is evident that something like one-fourth had passed through the cribriform plate into the lymphatics of the nose

Several studies have been made of the escape of particulate matter injected into the subarachnoid space under normal pressures, e g India ink (Brierley and Field, 1948), red blood cells (Simmonds, 1952, 1953) and thoracic duct lymph containing chylomicrons (Courtice and Morris, 1955) As far as red cells are concerned, the experiments of Simmonds (1953), using red cells labelled with P^{32}, point to a very slight removal *viâ* the nose and the cervical lymphatic pathway

The Termination of the Perineural Space

In all these studies concerning the passage of cerebrospinal fluid from the cranial subarachnoid space, one of the problems as yet unsolved is the mode of termination of the perineural space As the olfactory bundles pass through the foramina in the cribriform plate they are undoubtedly surrounded by an extension of the subarachnoid space, bounded on the outside by the dura and arachnoid and on the inside by the pial covering of the nerve Once outside the skull, the dura and arachnoid gradually approach the pial investment of the nerve, and finally appear to blend with it to form the perineural sheath or epineurium At the same time the dura-arachnoid appears to undergo progressive thinning The prolongation of the sub-arachnoid space along the olfactory nerve bundles clearly allows cerebrospinal fluid to pass along this wedge-like perineural space out of the skull into the submucous tissue of the nose The question not as yet resolved is how far this space extends, and what exactly are the structure and properties of the distal part of the dura-arachnoid If it is an intact membrane and if it is thin enough to permit the free passage of fluid, then the cerebrospinal fluid can readily traverse it, and once it is through it would readily be drained away by the submucous lymphatics If the perineural membrane is intact one must in fact postulate that this happens, or else it would not be possible to explain experiments such as those of Brierley and Field (1948), Simmonds (1953, 1952) and Courtice and Morris (1955), unless indeed the thinning dura-arachnoid disappears completely, in which case fluid in the peri neural space would drain directly into the submucous tissues

But if this latter is true, one would expect that substances in the sub mucous tissue could then pass back fairly readily into the interior of the cranium, and this does not appear to occur. If one leaves out of consideration living matter, such as viruses or bacteria, which present special problems of spread, it is evident that the movement of substances from the nose to the cranium does not occur, except in the case of simple crystalloids which can diffuse rapidly (Le Gros Clark, 1929; Faber, 1937-8; Yoffey and Drinker, 1938). Le Gros Clark seems to have been the earliest observer to demonstrate this fact. He placed a 0·5 per cent. solution of trypan blue in the nose of a rabbit for 24 hours, at which time he was unable to find any dye in the cranium. Yoffey and Drinker (1938) made similar observations with trypan blue and Evans blue which they left in the nose of cats, dogs and rabbits for several hours, while at the same time the deep cervical lymph ducts were being cannulated, and lymph from the

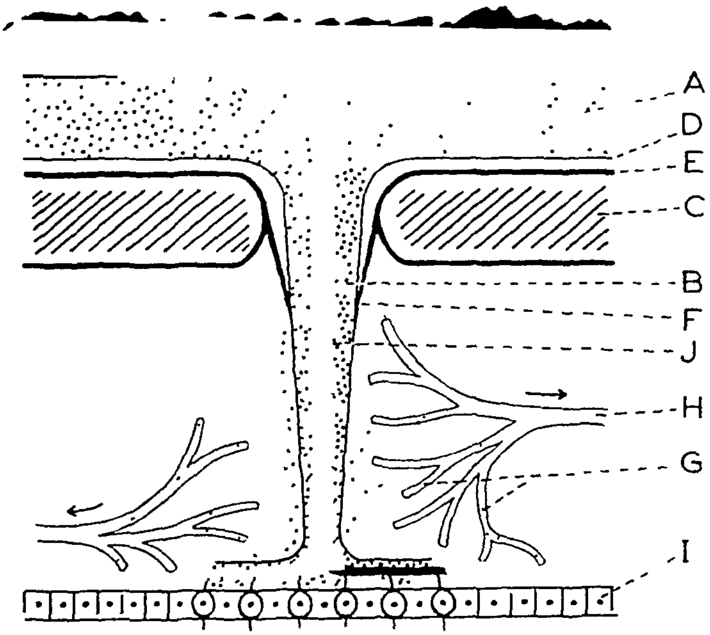

Fig 1

Schematic diagram to indicate the path taken by India ink particles from the cranial subarachnoid space to the lymphatics of the nose Modified from Brierley and Field (1948)

A = Subarachnoid space containing ink
B = Perineural space
C = Cribriform plate
D = Arachnoid
E = Dura
F = Fusion of dura and arachnoid with epineurium
G = Lymphatic capillaries
H = Lymphatic collecting trunk
I = Mucous membrane of nose
J = Olfactory nerve bundle

nose collected. Though dye was present in the lymph in high concentration for several hours, it, nevertheless, did not enter the cranium.

If the terminal part of the perineural sheath is intact, one can explain the failure of dye to enter the cranium either because it does not pass through the sheath, which functions as a semipermeable membrane, or because it passes through much more slowly than the outward current of cerebrospinal fluid. If the perineural sheath is incomplete, then the dye has free access to the perineural space, and the only possible explanation of its not reaching the subarachnoid space is the continuous flow of cerebrospinal fluid.

How far does the Perineural Space extend?

Reviewing the work of previous investigators, I think that in the case of some of the olfactory bundles, at least, the perineural space may extend practically as far as the olfactory epithelium (see for example Weed, 1914). Fig. 1 represents this schematically, and is modified from the diagram by Brierley and Field (1948). Any increase in pressure in the submucous tissues would then compress the epineurium against the nerve bundle, temporarily obliterating the perineural space, while at the same time it would drive fluid in larger amount through the endothelium of the lymphatic capillaries into the lymphatic vessels. This appears to be the simplest of all the explanations which have been put forward to account for the extreme difficulty with which substances pass from the nose to the cranium. The proximity to the nasal mucosa of the terminal parts of the perineural spaces would explain why substances, injected into the subarachnoid space under raised pressure, escape so readily through the nasal mucous membrane.

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POST-CRICOID CARCINOMA

By RONALD W RAVEN (London)

RADICAL surgical operations designed upon an accurate knowledge of the pathology of the disease have an assured place in the treatment of carcinoma of the hypopharynx For many patients it is the only treatment with the prospect of cure which can be offered to them Whilst this applies to carcinoma in other parts of the hypopharynx, it is especially relevant to the post-cricoid region My experience has convinced me that whenever possible a radical surgical operation should be carried out for these patients

Definition

The post-cricoid area is the part of the anterior wall of the hypopharynx which corresponds to the broad quadrilateral posterior half of the cricoid cartilage This is covered by the crico arytenoideus posticus muscle and the mucous membrane of the pharynx A carcinoma commences in this area and in the posterior wall of the hypopharynx opposite to it These tumours involve the pharyngo œsophageal junction which they may surround A carcinoma which arises in other parts of the hypopharynx such as the pyriform fossa and arytenoid region may extend downwards into the pharyngo œsophageal region and present the same treatment problems as the primary post-cricoid carcinoma In this discussion I will consider those carcinomas which involve the post-cricoid and pharyngo-œsophageal regions and exclude such tumours as those confined to the pyriform fossa and arytenoid areas

Special Features

The Paterson- Kelly Syndrome Mention is made of this precancerous condition in the post-cricoid region Changes occur in the mucous membrane and submucosa leading to webs, bands and strictures The condition is a chronic iron-deficiency state and other deficiencies, especially of vitamins B and C, may be present An impassable stricture may form necessitating surgical treatment, and I have performed a resection of the pharyngo-œsophageal region with an immediate end-to end pharyngo-œsophageal anastomosis for such a condition This may be a preventive operation for post cricoid carcinoma in these cases

Prognosis

The prognosis of carcinoma involving the pharyngo œsophageal junction treated by irradiation is bad and few patients have survived for five years. I feel it is essential that radical surgical treatment should be given an extended trial in these cases so that the end-results can be evaluated in an adequate series of cases over a period of 5 years. The results which have been achieved by surgery fully justify this attitude.

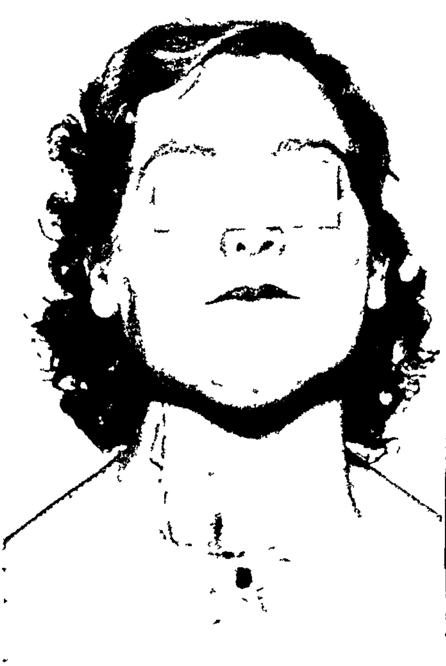

FIG 1

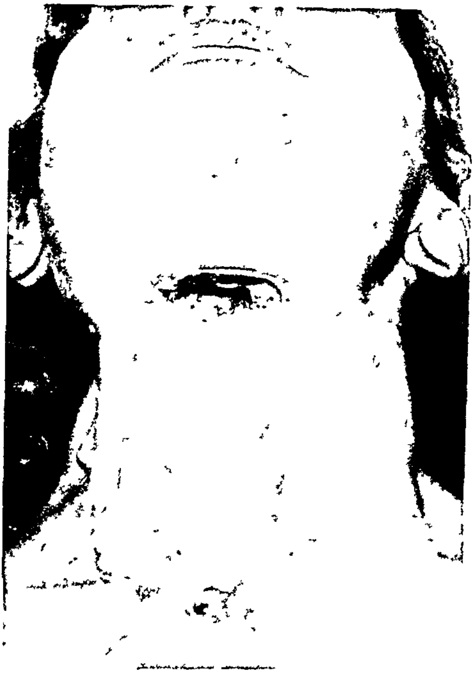

FIG 2

FIG 1

Photograph of a patient at the end of the first stage of laryngo-œsophago-pharyngectomy for post-cricoid carcinoma showing the œsophagostomy separated by a bridge of skin from the tracheostomy

FIG 2

Photograph of the same patient as in Fig 1 to show the pharyngostomy

Pathology in Relation to Radical Surgery

In some cases carcinoma in the hypopharynx is of multifocal origin. The tumour infiltrates in area and in depth in the pharyngeal wall so that other parts of the hypopharynx and structures in its vicinity are invaded. Upward spread occurs to the epiglottis and oropharynx and downward extension into the cervical œsophagus is common. Anteriorly infiltration into the larynx occurs and in an appreciable number of cases the wall of the trachea is penetrated In many cases there is direct spread into the lateral lobes of the thyroid gland and the internal jugular vein is frequently invaded and drawn anterior to the common carotid artery and becomes adherent to the pharyngeal wall

Metastases are frequent in the regional lymph nodes They form initially in the group of lymph nodes lying in close proximity to the internal jugular vein between the posterior belly of the digastric and the anterior belly of the omohyoid muscles, these nodes receive lymph from the hypopharynx through the supraglottic pedicle When the tumour is limited to one half of the hypopharynx the lymph nodes in both sides of the neck may nevertheless be affected These lymph nodes communicate with those lying along the spinal accessory nerve as far as the anterior border of the trapezius muscle and with others lying below the omohyoid muscle as far as the supra clavicular region

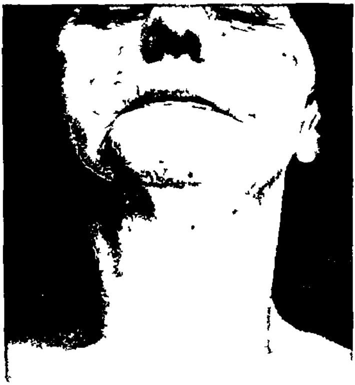

FIG 3
Photograph of a patient aged 61 with a large post cricoid carcinoma

A carcinoma in the post-cricoid region and cervical œsophagus causes metastases in the lymph nodes lying along the recurrent laryngeal nerves When the larynx is infiltrated metastases may be found in the prelaryngeal and pretracheal lymph nodes as well as in the perithyroidean lymphatics

A radical operation for a carcinoma in the hypopharynx must be planned and executed to include the removal of all the actual and potential affected tissues in the neck.

Operability

The selection of patients for operation is an onerous responsibility; when they are considered inoperable it is usually a death sentence. Alternatively a patient must not be submitted to radical surgical treatment when this is likely to leave him in a worse position later with an early recurrence or spreading carcinoma with the disadvantages of the presence of a pharyngostomy or œsophagostomy.

Clinical Assessment

The patient's physical condition must be suitable to withstand two major operations, and the cardiovascular, respiratory and urinary systems are assessed. Nutritional and blood deficiencies are estimated and corrected as far as possible. After a full course of radiotherapy the general condition is often poor and technical problems relating to the repair and healing of wounds must be solved.

The extent of the disease is assessed. Fixity of the hypopharynx or cervical lymph nodes renders the disease inoperable. In some patients the structures of the neck are incorporated as a malignant mass. Pharyngoscopy shows the level of upward extension which may reach the lower part of the oropharynx but the disease can still be removed. It is difficult to determine the extent of spread into the cervical œsophagus as an œsophagoscope cannot be passed in many cases. Information is provided by a radiological examination on swallowing barium and by tomograms which also give an indication of the general extent of the tumour.

When a post-cricoid carcinoma has spread into the thoracic œsophagus operation may still be possible by an extension of the laryngo-œsophago-pharyngectomy operation which includes excision of the manubrium sterni. The final decision whether the œsophageal extension precludes operation is usually made on exploration of the neck.

A lateral X-ray of the neck demonstrates the degree of involvement of the retro-pharyngo-œsophageal tissues and of the cervico-dorsal vertebrae.

Operative Assessment

The first stage in the radical operation is to determine the exact boundaries of the disease by inspection, palpation and the removal of tissues for immediate histological examination. The quadrilateral flap is raised and a thorough exploration of the neck is carried out. This includes an assessment of the extent of lateral spread by direct extension; involvement of the common or internal carotid arteries by the pharyngeal carcinoma renders it inoperable. A decision is made whether it is necessary to include a radical dissection of the cervical lymph nodes and on which side. The recurrent laryngeal lymph nodes are exposed on both sides and those in the supra-glottic pedicle. An immediate histological examination will determine the presence of metastases in the largest of these nodes. The jugulo-digastric lymph nodes are also critically examined.

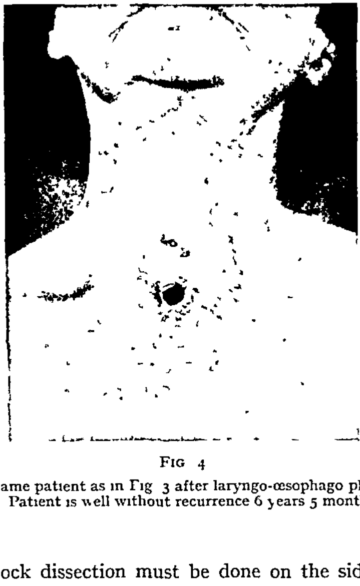

FIG 4

Photograph of the same patient as in Fig 3 after laryngo-œsophago pharyngectomy in two stages Patient is well without recurrence 6 years 5 months later

A radical block dissection must be done on the side which is more affected; when the lymph nodes on the other side are affected they must all be dissected out in the positions mentioned but the internal jugular vein is conserved.

The cervical œsophagus is exposed as far as the superior thoracic aperture and the lower level of the carcinoma is determined by palpation. When the œsophagus is divided later an immediate histological examination is made to be certain that the line of section has cleared the tumour edge. The same applies to the division of the upper end of the hypopharynx.

Choice of Operation

Three operations are considered for post-cricoid carcinoma. Partial œsophago-pharyngectomy with conservation of the larynx was done for one patient and a skin hypopharynx was constructed and anastomosed to the oropharynx above and the œsophagus below. The functional result was bad and the patient died with a recurrence after one year. This operation is not sufficiently radical and I do not advise conservation of the larynx in these cases. Sometimes a one-stage laryngo-œsophago-pharyngectomy can be performed with an immediate end-to-end anastomosis between the divided hypopharynx and œsophagus. This is an advantage because primary healing of the neck is obtained, but the anastomosis must not be done under tension.

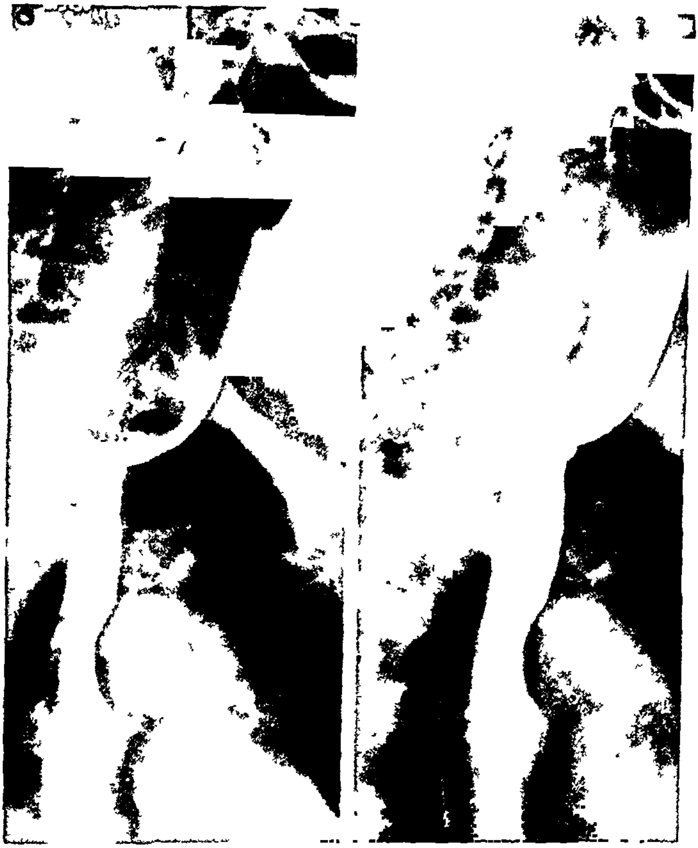

FIG 5

X-ray on swallowing barium of the new skin hypopharynx and cervical œsophagus of the same patient as in Fig 3 Antero posterior view

The best operation for post-cricoid carcinoma in my experience is a radical two-stage laryngo œsophago-pharyngectomy performed as a monoblock operation to include all the affected regional lymph nodes

Laryngo-œsophago-pharyngectomy in two stages. Technical considerations

I will stress those which I have found in my work to be important

Excision of affected regional lymph nodes a radical lymphatic dissection is performed on the side most affected and the dissection extends from the tip of the mastoid process above to the clavicle below, and from the anterior border of the trapezius muscle inwards The internal jugular vein is approached posteriorly and ligated first near the base of the skull and then below above the clavicle It is then dissected from the carotid sheath and left attached with the lymph nodes, sternomastoid muscles and the other tissues to the hypopharynx The affected lymph nodes are dissected from the opposite side from without inwards and left attached to the hypopharynx

The chain of lymph nodes lying along the recurrent laryngeal nerve are dissected out of the thoracic inlet on both sides and left attached to the pharyngo œsophageal junction

Division of the pharynx and œsophagus In all cases the pharynx is divided above the hyoid bone and the latter is removed with the hypopharynx The line of section must be at least 2·5 cm above the upper limit of the carcinoma, when doubt exists regarding clearance of the tumour a histological examination of the divided pharynx by the frozen section technique is made In cases with extensive upward spread of the carcinoma the section may pass through the middle part of the oropharynx near the base of the tongue The mucous membrane is then reflected off the back of the tongue and sutured to the submaxillary skin to form the anterior wall of the pharyngostomy The œsophagus is divided as low as possible in the thoracic inlet The cut end of the œsophagus is examined histologically by the frozen section technique to ensure perfect clearance of the tumour

Total thyroidectomy This is performed in every case, for tumour spread by direct extension into the gland is frequent and the perithyroidean lymphatic plexus is connected with the lymph nodes draining the post-cricoid region and the cervical œsophagus

Formation of the tracheostomy and œsophagostomy A quadrilateral pedicle flap of skin based on the inner end of the right clavicle is raised from the anterior aspect of the manubrium sterni and transposed to lie between the tracheostomy and œsophagostomy The anterior edge of this skin flap is sutured to the posterior wall of the trachea, the anterior wall of the latter is sutured to the skin over the manubrium sterni The anterior wall of the divided œsophagus is sutured to the posterior edge of the transposed skin flap, and the posterior wall of the divided œsophagus is sutured to the lower border of the main quadrilateral skin flap

Formation of the pharyngostomy. The anterior and posterior walls of the divided oropharynx laterally are sutured to diminish its diameter. The posterior wall of the divided oropharynx is sutured to the upper border of the quadrilateral skin flap; the anterior wall of the divided oropharynx or the mucous membrane reflected from the back of the tongue is sutured to the skin in the submandibular region.

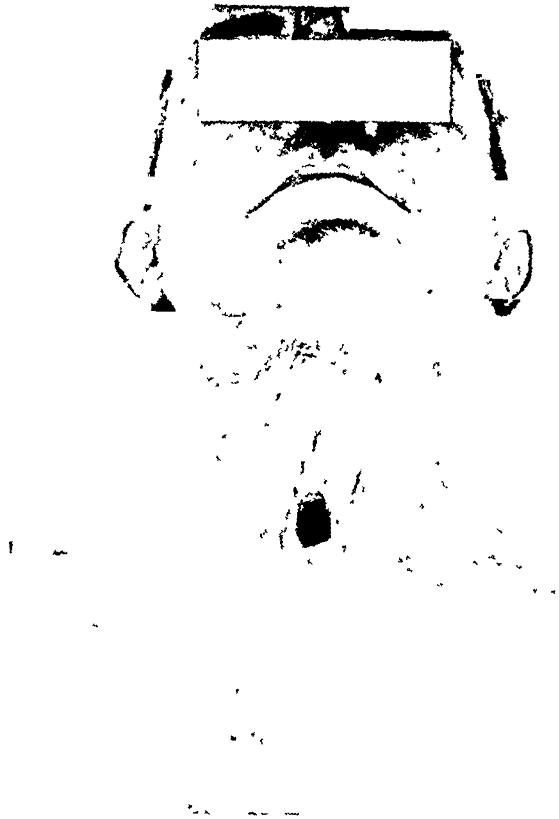

Fig 6.

Photograph of a patient, age 65, after completion of laryngo-œsophago-pharyngectomy in multiple stages for an advanced carcinoma of the hypopharynx uncontrolled by radiotherapy and which required a palliative tracheotomy Patient is well without recurrence 6 years 5 months later

The free edge of the quadrilateral skin flap is sutured to the skin of the opposite side of the neck.

Interval between the operation stages. I usually allow an interval of eight weeks to elapse before the pharynx is reconstructed so that the skin to be used for this purpose acquires an adequate blood supply from the prevertebral region.

Reconstruction of skin pharynx and cervical œsophagus. The following technical points are important. An incision is made through the healed longitudinal incision in the neck from the oropharynx to the side of the tracheostomy, and a second incision is made on the opposite side of the neck. These incisions are convergent and the outer parts of the new pharyngeal wall are reflected inwards so as to form a tube. The anterior wall of the pharyngostomy is separated from the skin of the submandibular region and sutured to the upper edges of the new skin tube.

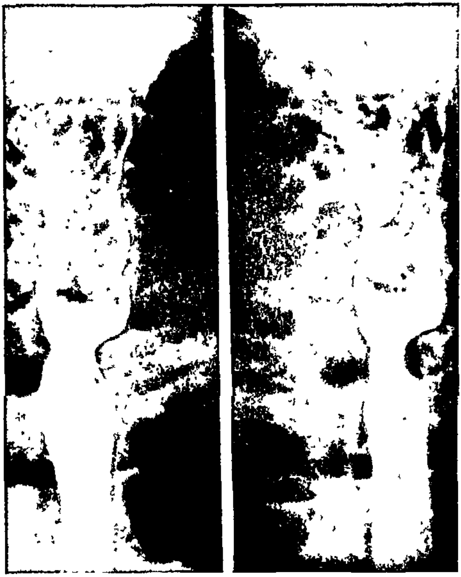

FIG 7.

X-ray on swallowing barium of the new skin hypopharynx and cervical œsophagus of the same patient as in Fig 6

The œsophagostomy is separated from the tracheostomy by dividing the skin joining the posterior wall of the latter structure. The skin flaps are then mobilized and sutured together and the rest of the anterior wall of the new skin pharynx is closed by interrupted sutures.

Skin cover for new skin hypopharynx and cervical œsophagus. A flap of skin is reflected from the postero-lateral aspect of the neck to cover the new skin pharynx and œsophagus. This is based on the occipito-mastoid region. Any raw area left is covered by a split-thickness skin graft.

Radical Surgery following Radiotherapy

The vitality of the tissues of the neck are often impaired by a full course of radiotherapy and difficult technical problems are created when radical surgery is required for an *uncontrolled carcinoma in the post-cricoid area*. I have found the following techniques of value in dealing with these patients.

Ensure viability of the quadrilateral skin flap which is required for the posterior wall of the skin hypopharynx and cervical œsophagus. The flap must be fashioned as narrow as possible and not reflected off the sternomastoid muscle at its base. It is essential *to achieve good vitality of this* posterior wall and primary healing of the anastomosis.

Obtain cover for the carotid arteries. As radio-necrosis of the skin and superficial tissues may occur, the wall of the carotid arteries, which may be already weakened, must be protected. On one side the remaining sternomastoid muscle is sutured to the prevertebral tissues and provides good cover. It may be possible to conserve the other sternomastoid for the same purpose and this should be done whenever possible otherwise an area of radio-necrosis may occur over it and fatal hæmorrhage can result. Adequate skin cover for the carotid arteries *must always* be provided.

Provision of skin for the anterior wall of the new skin hypopharynx and cervical œsophagus. This is obtained from the anterior chest wall by acromio-pectoral tubed pedicle grafts which are transplanted in stages to the neck. The condition of the skin and other structures in the neck improves when an additional blood supply is furnished by the tubed skin grafts. Multiple stages over a long period of time are required before the patient is able to swallow perfectly; I have taken up to two years to complete these operations.

Results of Radical Surgery

The results obtained in a series of 37 cases with carcinoma of the hypopharynx treated by radical surgical operations are shown in Table I. The survival periods are computed in months from the date of the first excisional operation for the carcinoma.

TABLE I

CARCINOMA OF HYPOPHARYNX. RESULTS OF RADICAL SURGERY. SURVIVAL IN MONTHS AFTER OPERATION

Number alive	15	2	2	2	3	8	12	15	19	21	27	57	60	77	77	78
Number died of cancer ..	15	4	4	6	8	8	12	12	12	15	15	20	23	23	33	66
Number died of other cause ..	4	2	10	30	91											
Number died of operation ..	3	3 days			23 days			6 weeks								
Total Number	37															

The results must be judged against this background The majority of these patients treated by radical surgery had advanced carcinoma in the hypopharynx for the tumour had affected extensive areas and frequently infiltrated the cervical œsophagus, larynx, trachea and thyroid gland In 16 patients (47 per cent) there were metastases in the cervical lymph nodes which were proved by histological examination The patients suffered severely from dyspnœa, stridor and dysphagia, two patients had a palliative gastrostomy and another a palliative tracheotomy Some had been pronounced untreatable and were awaiting death with the help of sedatives Others had received radiotherapy but the disease was uncontrolled The only encouraging feature was the splendid spirit of the patients and their desire that something should be done to help them in their plight It is pointed out that a radical operation provides immediate palliation in addition to giving the possibility of cure

Three patients died at intervals of 3 days, 23 days and 6 weeks after the operation The causes of death were respectively cerebral anoxia, bronchopneumonia and carotid artery hæmorrhage

Four patients died of causes other than cancer, the longest survivor in this group lived 7 years 7 months without recurrence of the carcinoma and she died age 75 years from arteriosclerosis and hypertensive heart failure

Fifteen patients died of cancer at intervals varying from 4 to 66 months after the excisional operation The longest survivor in this group died from a carcinoma of the tongue

Fifteen patients are alive at intervals varying from 2 to 78 months after the excisional operation The numbers of survivors at different intervals following the excisional operation are shown in Table II

TABLE II

CARCINOMA OF THE HYPOPHARYNX NUMBER OF SURVIVORS AT DIFFERENT INTERVALS FOLLOWING THE EXCISIONAL OPERATION

Interval in Months	Number of Survivors
12	22
24	10
36	7
48	7
60	6

In 14 patients in this series the carcinoma commenced in the post-cricoid region or invaded it from some other primary site in the hypopharynx Seven patients were alive 1 year after the operation, three patients were alive more than 5 years later, namely for 77, 77 and 91 months respectively

These surgical operations do not cause mutilation, the patients have a normal neck except for healed incisions and a tracheostomy. They can dress normally and swallow everything, and I have not had a case of fibrous stricture formation at the junction of the skin pharynx and cervical œsophagus or elsewhere. The patients are able to work and to live almost normal lives.

Note: Since this paper was given in June, 1957 three of the patients in Table I have died of cancer 8, 9 and 13 months after the operation, leaving 12 survivors. The longest survivor in this group is alive in April, 1958, 88 months after the operation.

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POST-CRICOID CARCINOMA

By M LEDERMAN (London)

IN the absence of any agreed classification, it is still necessary to define the term "post-cricoid cancer", otherwise there can be no certainty that the same tumours are being discussed by different speakers The term "post-cricoid" is time-honoured and widely used, but it refers not so much to a definite anatomical site or a single clinical form of cancer as to the region lying behind the larynx where neoplasms may occur and can be detected by mirror examination The common tumours arising in this so-called post-cricoid region can be subdivided into three main groups

(*a*) Cricopharyngeal
(*b*) Pharyngo-œsophageal
(*c*) Cervical œsophagus

(1) Crico-pharyngeal Tumours

This group includes all the tumours arising from the regions usually designated "post-cricoid" or "mouth of the œsophagus" Tumours of these parts may arise anteriorly from the mucosa overlying the posterior surface of the cricoid or crico-arytenoid region, or posteriorly from the mucosa covering the cricopharyngeus, some lesions may arise laterally from the lowest part of the pharyngo-laryngeal grooves

(2) Pharyngo-Œsophageal Tumours

This term can be applied to tumours arising from the region of the œsophagus just below the crico-pharyngeal sphincter Characteristically, these tumours affect equally both the lower pharynx and upper œsophagus and the thyroid gland is also often invaded, the resulting clinical picture being frequently one of a patient who can neither swallow nor breathe

(3) Cervical Œsophagus

Whilst it may seem somewhat out of place to include tumours of the cervical œsophagus in this discussion, such a step can be justified both on diagnostic and technical grounds Advanced œsophageal lesions do present in the post-cricoid region and their true origin and extent may be overlooked in the absence of thorough clinical and radiological examination As far as surgical or radiological treatment are concerned, the œsophagus cannot with safety be divorced from the lower pharynx and it is worth noting that a great deal of the surgical literature concerns itself as much with the "upper œsophagus" as with the "post-cricoid region". Jacobsson (1951) notes that two-thirds of the cancers affecting the lower pharynx extend into the œsophagus and 45 per cent. of the tumours are more than 5 cm. long.

Radiotherapy Technique (1956)

In discussing the technique of radiotherapy, the important point to establish is that 200 kV. X-radiation is the least desirable method of treatment, the harder quality gamma radiation from radium or cobalt, and supervoltage X-radiation being preferable. By the latter methods, higher doses can be given with less risk of complications and with the minimum of discomfort to the patient. Of far more importance to the radiotherapist than the possession of special radioactive isotopes or modern supervoltage equipment is his ability to use a laryngeal mirror and to individualize treatment. A standard tumour does not exist nor is there any special tumour dose, the administration of which is likely to be regularly successful. In these circumstances, the best chance of success does not lie in forcing the patient to do battle with a prescribed "lethal tumour dose" of radiation, but in individualizing treatment according to the patient's needs. To this end, regular and frequent examination of the patient by the radiotherapist is essential. The radiotherapist who cannot or will not examine the larynx regularly during the course of radiation treatment fails in his duty and may well reduce his patient's chances of success as well as add to the risks of treatment.

At the Royal Marsden Hospital there are two small telecurie therapy units; one, containing 12 gm. of radium, has been in constant use with variable quantities of radium for 21 years, and a second containing 10 curies of Cobalt 60 which has been available for the past 5 years. For all practical purposes, the radium and Cobalt 60 can be regarded as equivalent in their effects and uses.

The technique of treatment depends on the age, general condition of the patient and the site and extent of the tumour. The anæmic, dehydrated and emaciated patient is a poor subject for radiotherapy, but it is very rare to have to delay treatment on these grounds. Occasionally nasal feeding is necessary or even a gastrostomy may have to be performed but generally, if the patient's condition is so bad that a gastrostomy or tracheotomy is necessary, then it is often worthwhile offering radiotherapy. 20 per cent. of our patients had to have a gastrostomy, 10 per cent. a tracheotomy and 5 per cent. had both gastrostomy and tracheotomy.

As a general rule, telecurietherapy is used for the earlier cases, X-ray therapy being reserved for advanced cases requiring palliation. The treatment fields used depend on the part of the pharynx or œsophagus affected but in all cases a wide segment is irradiated and the appropriate lymph node areas must be routinely included in the treatment area

The doses employed vary using telecurietherapy the aim is to give 6,000-7,000 r in 6-8 weeks, and using 200 kV X-rays, 4,500-5,500 r in the same time

Material and Results

During the period 1933-1956, some 623 cases of laryngopharyngeal cancer have been seen at the Royal Marsden Hospital (Table I) Much of this material has come during the last 8 years as a result of our co operation scheme with the Royal National Throat, Nose and Ear Hospital, and more recently with the Metropolitan E N T Hospital Table II lists the post-cricoid cases in relation to site of origin and stage, the basis for staging being the condition of the lymph node areas of the neck and the presence or absence of laryngeal mobility (Lederman, 1954) At least two-thirds of the cases come into the advanced stages III and IV

TABLE I

CA LARYNGOPHARYNX 1933 1956

MATERIAL

Group	Number Seen	Previously Untreated	Recurrent or Post operative	Not treated by Radiotherapy
Epilaryngeal	132	119	8	5
Sinus Pyriformis	234	208	16	10
Posterior and lateral walls	35	28	7	—
Post Cricoid Cricopharyngeal Pharyngo œsophageal Cervical œsophagus	222	94 64 = 197 39	19	6
Total	623	552	50	21

TABLE II

POST CRICOID CANCER 1933 1956

DISTRIBUTION ACCORDING TO SITE AND STAGE

Site	Stage				Total
	I	II	III	IV	
Cricopharyngeal	9 (10%)	23 (24%)	40 (43%)	22 (23%)	94 (48%)
Pharyngo œsophageal	6 (9%)	12 (19%)	27 (42%)	19 (30%)	64 (32%)
Cervical œsophagus	4 (10%)	13 (33 3%)	9 (23%)	13 (33 3%)	39 (20%)
Total	19 (10%)	48 (24%)	76 (38%)	54 (27%)	197

The results of treatment are notoriously bad and our results are no exception. From Table III the points of interest which emerge are:

(1) The Stage I cases which would be of most interest to the surgeon, form the one group which gives a worthwhile result following radiotherapy. It is salutary to reflect on the paucity of the really early case—19 in a 24-year period!

(2) The results obtained in selected cases of carcinoma of the cervical œsophagus merit special attention because of the particularly poor surgical results in this group.

(3) The results in the remaining groups are deplorable and are in fact the worst for all the laryngopharyngeal tumour sites.

TABLE III

POST-CRICOID CANCER: 1933-1956

RESULTS ACCORDING TO SITE AND STAGE

Site	5-year results: 1933-1951					3-year results: 1933-1953				
	I	II	III	IV	All	I	II	III	IV	All
Cricopharyngeal	2/7	0/10	2/24	1/15	5/57 (9%)	3/7	1/13	1/29	1/16	7/65 (11%)
Pharyngo-œsophageal ..	1/5	0/6	0/17	0/15	1/43 (2%)	2/6	0/9	2/20	0/15	4/50 (8%)
Cervical œsophagus ..	3/4	1/4	0/3	0/4	4/15 (27%)	3/4	2/5	0/6	0/8	5/23 (22%)
Total	6/16	1/20	2/44	1/34	10/115 (9%)	8/17	3/27	4/55	1/39	16/138 (12%)

No one can complacently accept results of this kind: in the search for future improvement there are the following possibilities to consider:

(1) Developments in radiotherapeutic apparatus and technique. There would appear to be an optimum range of energy of radiation (1-5 MeV) for the treatment of head and neck cancer. Many centres now have apparatus in routine use capable of providing radiant energy of this quality and there is, as yet, no evidence to show that either newer or more powerful equipment or the use of more esoteric techniques or isotopes will materially influence the prospects of the patient suffering from post-cricoid cancer. The reason for this pessimistic outlook rests on the fact that radiation is a non-specific remedy and therefore such sporadic radiocurable cases as are encountered should be curable by existing equipment and technical methods.

This same argument applies to:

(2) The possibility of developments in surgical technique. The natural

history of the disease and the non-specificity of the remedy militate just as much against the surgeon as the radiotherapist. There is no lack of boldness or technical skill among present-day surgeons, yet as a rule the greater the surgical effort the less the reward. If the prospects for both surgery and radiotherapy used independently are admittedly poor, can (3) radiotherapy and surgery in combination offer more? As the results could scarcely be worse, this approach must be seriously considered. It is the writer's view that there may well be a place for some combination of surgery and radiotherapy in the treatment of post-cricoid cancer but the precise manner of combination is of vital importance.

It is natural that the surgical part of the treatment should be considered first since the patient is rapidly relieved of most of his neoplasm and the difficulties and complications of excision after radiotherapy are avoided: also a number of patients who receive radiotherapy first and improve, may well be tempted to refuse any subsequently proffered operation. In spite of this, however, the fundamental differences between pre- and post-operative radiotherapy must be appreciated and considered by both surgeon and radiotherapist before deciding on any particular combination of methods.

Where radiotherapy is employed *before* operation, its purpose is to assist the surgeon in his task and this is achieved by shrinkage of the tumour, diminution of sepsis and reduction in viability of tumour cells so that the likelihood of recurrence in the operative field is lessened. These benefits can be conferred by radiotherapy without necessarily employing *full dosage or unduly delaying the operation.*

The purpose of post-operative irradiation is entirely different since radiotherapy is of no prophylactic value in an immunological sense and, therefore, if a tumour has been completely removed its use is pointless since future recurrence or metastasis cannot be prevented by this means. If, however, removal has been incomplete, then the purpose of radiotherapy is to salvage the patient and a full course of treatment becomes necessary. In these circumstances, even when full doses have been used, radiotherapy is still not very effective since cancer cells present in scar tissue rarely respond satisfactorily and treatment as a rule proves only of palliative value.

In the writer's opinion, if surgery and radiotherapy are to be combined to the patient's advantage, radiotherapy should be employed first and then followed by surgery. Obviously each case would have to be considered individually: in the advanced, complicated case where a poor response to radiotherapy was shown, there would be little point in persevering beyond a dose of 2,000 r in 2-3 weeks. When, however, a favourable response is observed treatment could be continued and full dosage given in the hope of obtaining a cure by radiotherapy alone. Provided that hard quality radiation is used and repeated courses of treatment avoided, the problems of surgical removal and reconstruction, although aggravated by the previous course of radiation, need not prove insoluble.

Table IV lists the surgically treated cases in this series.

TABLE IV

POST-CRICOID CANCER

OPERATIONS 1933-1953	
No. performed	4
Lateral pharyngotomy and block dissection	1*
Pharyngo-laryngectomy	2
Transthyroid pharyngectomy	1

* Patient lived 5 years. Other patients died of disease within 2 years.

1954-1956 (INCLUSIVE)	
No. performed	5
Laryngopharyngectomy	2
Laryngopharyngectomy and block dissection	3

1 patient is alive for 1 year
4 patients died within 1 year

(4) Prevention

Logically, prevention would provide the only satisfactory solution to the problem of post-cricoid cancer—or any form of cancer for that matter. Fortunately a study of these cancers shows without doubt that certain of them possess features of such ætiological and clinical significance that the possibility of prophylaxis must be considered seriously. Thus:

Laryngopharyngeal (hypopharyngeal) cancer is common in most European countries but there is a remarkable geographical variation in the frequency with which the post-cricoid region is affected and the frequency with which women are the sufferers. In Table V the sex incidence of laryngopharyngeal cancer in different countries is well shown. It would

TABLE V

HYPOPHARYNGEAL (LARYNGOPHARYNGEAL) CANCER.
SEX INCIDENCE IN DIFFERENT COUNTRIES (AFTER JACOBSSON)

Country	Percentage of women
France (Baclesse)	1
Switzerland (Schinz & Senti)	9
U.S.A. (Martin)	15
Denmark (Videbaek)	20
England (Cade)	25
Finland (Sipila)	30
Ireland (Douglas)	40
Norway (Rennaes)	50
Scotland (Turner)	>55
Sweden (Radiumhemmet)	60
Royal Marsden Hospital	31

seem that in the Latin countries and Switzerland (Jacobsson, 1951, Leroux-Robert, and Ennuyer, 1952, Agazzi, 1953), women rarely develop laryngopharyngeal cancer, men being the chief sufferers and the pyriform sinus the region usually affected There is also a geographical variation in the frequency with which the post-cricoid region is affected (Table VI), the site being commonly affected in Scandinavia, whilst it is excessively uncommon in France and Italy and the Mediterranean countries generally (Leroux-Robert and Ennuyer, 1952, Agazzi, 1953)

TABLE VI

INCIDENCE OF POST CRICOID TUMOURS RELATIVE TO OTHER LARYNGOPHARYNGEAL SITES (AFTER JACOBSSON)

Country	Per cent Carcinoma of lower part of hypopharynx*
England	25
Finland	25
Ireland	35
Norway	40
Scotland	50
Sweden	75
Royal Marsden Hospital	30

* post cricoid region

It would appear, therefore, that in the Latin countries it is the male who develops laryngopharyngeal cancer, usually in the sinus pyriformis, whilst in the Scandinavian countries it is the female who develops this form of cancer and usually in the post-cricoid region In the Anglo-Saxon countries the incidence seems to fall between these two extremes It should also be noted that irrespective of country, when a woman develops laryngopharyngeal cancer it is the post-cricoid region that is most likely to be affected

As an explanation of the extraordinarily high incidence of laryngopharyngeal cancer in women in Sweden, Ahlbom (1941) pointed out that 90 per cent of these women showed signs of sideropenic dysphagia, i e the Plummer Vinson syndrome and simple achlorhydric anæmia Apart from sideropenia, there may be other characteristic clinical features such as rhagades at the corners of the mouth, atrophic changes of the buccal and lingual mucosae, and changes in the fingernails (Lindvall, 1953) The fully established picture originally described by Ahlbom has rarely been seen by the writer, instead, many patients show some, but not all, of these features to a greater or lesser degree

In Table VII it can be seen that this syndrome is much less common in this country than in Sweden Of 106 patients especially examined,

TABLE VII

SIDEROPENIC DYSPHAGIA (AHLBOM'S SYNDROME)

Number of patients previously untreated			197
Changes noted		60	
Tongue smooth, mm. atrophic or smooth	3		
Fissures at corners of mouth, nail changes	4		
Tongue smooth, fissures at corners of mouth	12		
Fissures at corners of mouth	9		
Tongue smooth, nail changes	10		
Nail changes	8		
Tongue smooth	14		
No changes noted		46	
Not recorded		91	
		197	

BLOOD COUNT

Number of patients previously untreated		197
Blood count within normal limits (Hb. less than 70 %)	127	
Low blood count (RBC less than 4,000,000)	14	
Not recorded	56	
	197	

DYSPHAGIA

Number of patients previously untreated			197
No. of patients with dysphagia as first symptom		139	
Dysphagia present over 1 year	28		
Dysphagia present over 5 years	17		

60 showed some changes of the tongue and oral mucosae and nails. Of 127 patients whose blood count is recorded, only 14 (11 per cent.) showed any significant change and of the 139 patients in whom dysphagia was the dominating symptom, only 17 (12 per cent.) had a long history that might be suggestive of Plummer Vinson's syndrome.

Bearing these facts in mind, a possible manner of prevention may well be along the following lines:

(1) Detection by mass survey. This could follow the example of other mass surveys for the detection of malignant disease, e.g. gynæcological cancer. The investigation could be restricted in the first instance to women in certain age groups who admit to symptoms suggesting anæmia, avitaminosis or difficulties in swallowing. The routine initial examination would include special attention to the state of the mucous membranes of the mouth and pharynx, the nails and teeth, and a blood examination. It should be possible to detect patients who present clinical features suggestive of Plummer Vinson syndrome or so-called sideropenic dysphagia

CARCINOMA OF THE HYPOPHARYNX IN THE FEMALE

By R. G. MACBETH (Oxford)

CARCINOMA of the hypopharynx in the female is not a common disease. In the ten-year period 1948-57, only 34 cases have been sent to the Department of Otolaryngology at Oxford, which has an average annual attendance of 8,000 new out-patients.

The majority of these cases (27) belonged to the group of true post-cricoid carcinoma, originating in women who had previously suffered from microcytic anæmia. In these the prognosis appears to be poor whatever is done, because of early metastasis and must be regarded as hopeless where metastases are clinically evident.

A small minority (5) belonged to a quite distinct group, where the disease originated on the posterior or lateral wall of the pharynx at about the cricoid level. These were otherwise healthy women who had never been anæmic and their prognosis was very much better than that of the post-cricoid group.

The choice of method of treatment traditionally lies between a pharyngo-laryngectomy and irradiation, with reliance upon surgical salvage of the failures of irradiation and irradiational palliation of the failures of surgery.

I have come to the following conclusions based upon my own experience and the experiences of others:

(1) A pharyngo-laryngectomy is a major operation which curtails function. In order to be successful as the primary treatment it must be carried out on the early case.

(2) Primary reconstructive grafting carried out at the time of a pharyngo-laryngectomy—whether in irradiated tissue or unirradiated—leads too often to stricture formation for it to be regarded as a satisfactory surgical technique. Therefore a two-stage operation is to be preferred.

(3) Deep X-ray therapy seems to be rarely successful in post-cricoid carcinoma; but it checks metastasis.

(4) Deep X-ray therapy ruins the tissues of the neck from the point of view of subsequent radical and reconstructive surgery.

These conclusions posed a problem whose solution has, I hope, been aided by the interstitial use of radioactive tantalum wires wrapped around the pharynx and larynx, and I have to thank Dr Frank Ellis for permission to mention the method here This material is introduced inside fine polythene tubing at an open operation conducted through a midline incision The polythene is brought out either through punctures in the skin or through the midline incision A tumour dose equivalent to 4,000 r is usually given

This method must be regarded as essentially experimental, but it appears to have produced one short term primary cure in a post-cricoid case and two in posterior wall cases and to be leaving neck skin which can be used for later pharyngeal reconstruction if the need arises

The actual method of pharyngo-laryngectomy, with a "gutter graft" on the prevertebral fascia as suggested by Eric Peet has been described already * It is usually possible to carry out a secondary closure about three weeks later

Conclusions

It is impossible to draw firm conclusions from a small series, but the figures seem to show that where radical surgery is carried out at some stage of the treatment the survival expectation is better than where irradiation only is employed

I have the feeling that the majority of these patients will prove to need both irradiation and surgery and perhaps we should deliberately plan this I think that interstitial irradiation has many advantages over teleradiation and because it may cure a few cases it should precede surgery Finally, a two-stage operation is much more likely to provide a satisfactory swallowing tube than is a one-stage operation

HYPOPHARYNGEAL CARCINOMA IN FEMALES 1948 57

Diagnosis		Dead	Alive	Untraced
True post cricoid	27	21	5	1
Posterior pharyngeal wall	5	2	3	—
Carcinoma of thyroid	2	2	0	—
TOTAL	34	25	8	1

CAUSES OF DEATH IN HYPOPHARYNGEAL CARCINOMA

Cancer	20	(with secondaries in 19)
Operation or sequelae	3	
Other	2	
	25	

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* Macbeth R G 1954 *Irish Jnl med Sci* 141

Methods of Ttreatment

	P.C.C.	Alive	P.W.	Alive
Surgery only	5	1	1	0
Surgery followed by irradiation ..	5	0	—	—
Irradiation only (*a*) D.X.R. ..	2	1	2	1
(*b*) Tantalum implant	4	1	2	2
D.X.R. followed by surgery ..	1	0	—	—
Tantalum followed by surgery	2	2	—	—
No treatment	7	0	—	—

Post-Cricoid Carcinoma Survival (Years)

Method	<1	1-2	2-3	3-4	4-5	5
Surgery only	4				1	
Surgery + Irradiation	2	2	1			
Irradiation only (*a*) D.X.R. ..	1	1				
(*b*) Tantalum implant	3	1	1			
Irradiation + Surgery	1		1			
No Treatment	7					

Pharyngeal Wall Survival (Years)

	<1	1-2	2-3	3-4	4-5	5
Surgery only			1			
Irradiation only (*a*) D.X.R. ..		1				1
(*b*) Tantalum Implant	1		1			

POST-CRICOID CARCINOMA

By J A SEYMOUR-JONES (Portsmouth)

THIS short paper indicates the incidence of post-cricoid carcinoma in the Portsmouth Hospital Group area over a ten-year period from 1947 to 1957

The Portsmouth Group is compact, consisting of the City of Portsmouth and the neighbouring townships of Gosport and Fareham, together with certain rural hamlets and small towns, the total population being estimated at about 350,000 There was dense overcrowding before the war, but the housing estates built since have absorbed a considerable proportion of the population Heavy industry is provided by Her Majesty's Dockyard and allied establishments There are a number of factories engaged in light industry Members of Her Majesty's forces form an appreciable group and are excluded as they are treated in Service hospitals A large number of elderly and retired persons also reside in the area, the climatic conditions being good and the atmosphere remarkably free from smoke Several cases have been included from the Chichester area, which is visited by one of the Portsmouth laryngologists

After the inception of the National Health Service, all specimens for histological examination were referred to the Portsmouth Group Central Laboratory and the Director of the Radiotherapy Department, Mr. Gunderson, was invariably consulted at an early stage on the advisability of radical surgical intervention and the radio-therapeutic management

The number of cases collected is 41, of which the majority were over 65 years old, 15 males and 16 females, but there was a small group of young persons, 1 male and 3 female

It is considered that these cases comprised a great majority, but not all, of those arising in the area served by the Group Hospitals It is also felt that all were genuine cases of post cricoid carcinoma, although in very few instances the tumour might be more properly regarded as having arisen in the pyriform fossa It is not always easy to be certain in more advanced cases of the precise origin of the growth Biopsy made in all cases revealed squamous-celled carcinoma in various stages of differentiation All cases treated by radiation received high voltage therapy and none have been subjected to radio-active cobalt In the 25-35 age group, 1 male, 2 female 35-45 group, 1 female 45-55 group, 3 male, no female 55 65 group, 2 male, 1 female Over 65, 15 male, 16 female Of those who received high voltage X-ray therapy alone, 2 were alive and well at the end of the five-year period. The prospects of survival of cases treated by high voltage radiotherapy being on the whole poor, it was decided to subject cases considered suitable to radical surgery. These received pharyngo-laryngectomy with block dissection of the side of the neck on which enlarged glands were palpated. No cases were found suitable for conservative surgery, the larynx on examination afterwards being invariably invaded by the growth. In the first case after pharyngo-laryngectomy a primary Thiersch grafting was performed over a portex tube triangular in section and satisfactory post-operative swallowing was obtained, although the patient died in just under two years with recurrences.

In the second case an elderly woman, who had been sent back from Africa as inoperable with a gastrostomy, mediastinitis had already taken place in the perforation of the growth. It was therefore thought advisable to perform the Wookey type of operation. After the first stage a nodule appeared in the middle of the large skin flap, which was excised and proved to be malignant. The second stage was thereafter carried out with good immediate results. Because of the possibility of unsuspected malignant deposits being present in the wound, it was considered that the second-stage operation of Wookey, despite increased hospitalization, might be preferable and therefore the subsequent cases were all operated on by that method. It was not possible to complete the second stage, that is the reformation of the pharynx by skin flaps, in several cases because of the frailty of the patient. In one case, a man of 57, a certain amount of cutaneous breakdown occurred, which might have been averted by more adequate immediate post-operative splinting of the patient's head and neck. This case has since been treated by the plastic surgeons with two-layer skin flap coverage of the small fistulae which remained and a good result has been achieved.

Conclusions can be drawn from the radical operative treatment by Wookey's method, admittedly in a small number of patients (12 in all). With the modern methods of anæsthesia, supplemented by maintenance of circulating blood-volume as required, patients of any age appeared to stand even extensive and prolonged surgery extremely well. There was one anæsthetic death, the cause of which was known and could have been avoided, otherwise immediate mortality was nil. Serious post-operative stenosis did not occur except in one case later on from malignant deposits in neck gland. The second stage closure is relatively simple associated with very little or no shock. It is important to provide good fixation of the head and neck by splintage, especially after the first stage to avoid any breakdown of the skin flap and subsequent infection and scarring, which makes the subsequent closure much more difficult. Even after what appeared to be adequate radical surgery, malignant deposits in the neck glands tended to appear at varying intervals post-operatively. Spread may also occur directly and by blood and lymphatic channels, as is usual with carcinoma. High voltage X-ray of the neck glands proved to be only palliative, the growth in general being extremely radio-resistant. Disease in the small group of young persons encountered appeared to advance very rapidly and appeared to be surgically intractable. The tumour was invariably well advanced when first seen. Apart from the group of young persons already mentioned, all cases operated on gained weight and some were able to return home swallowing for a period of comparatively pleasurable existence. They were also relieved of malignant ulceration of the hypopharynx which causes great distress when untreated. On the whole, it is felt that the results are disappointing and we are as yet unable to record a single five-year cure, but the work is not entirely unrewarding. It may well be that one one-stage operation is preferable, but, though the disease is not extremely rare, it is not often present in a form amenable to surgery and experience is hardly gained.

CLINICAL RECORDS

RHINOSCLEROMA AND MALIGNANCY

TWO CASES OF RHINOSCLEROMA ASSOCIATED WITH CARCINOMA

By O. M. ATTIA* (Cairo)

RHINOSCLEROMA is a chronic granulomatous condition which affects mainly the nose and nasopharynx and is fairly common in Egypt. Its ætiology, clinical features, pathological picture and methods of treatment have been discussed by many authors. It is known that this granuloma spreads mainly in the soft tissues and in the connective tissue spaces and that it does not invade bony structures. It is not recorded that rhinoscleroma assumes malignant characters or predisposes to malignant change.

I am going to report here two cases of typical rhinoscleroma associated with an epidermoid carcinoma. When I saw the first case in October, 1953, I hesitated to assume a causal relationship between the two lesions as it was the first instance amongst several cases examined both clinically and pathologically. Therefore, at that time I thought that the two diseases might have occurred together by mere coincidence. But when I came across the second case one year later, in a routine pathological examination of a case of rhinoscleroma without any malignant change being suspected clinically, the question was again raised. The two lesions might not be present together by mere coincidence but the chronic irritation and sepsis produced by rhinoscleroma and which, as we know, lead to squamous metaplasia of the covering nasal mucosa, might in the long run lead to malignant change in this epithelium.

Case Reports

Case I. A male patient, aged 40 years was admitted to Demerdash hospital with a huge ulcer affecting most of the bony and cartilaginous parts of the nose and complicated with secondary infection. The clinical diagnosis was either a specific or a malignant ulcer and a biopsy taken. This was examined and the report (567/53) issued at that time read: "The biopsy received shows the presence of two pathological lesions:

(1) A typical rhinoscleroma with numerous Mickulicz cells and some Russell bodies partly covered with stratified squamous epithelium.

(2) A moderately differentiated squamous-cell carcinoma which is seen infiltrating the tissues of the rhinoscleroma and probably related to the covering epithelium (Fig. 1).

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* Pathology Department, Abbassia Faculty of Medicine, Cairo.

It would be of interest to know the previous history of this case and whether it started as a granuloma (rhinoscleroma) which was later on complicated by a malignant ulcer or whether the two lesions are present together just by mere coincidence. Was the presence of rhinoscleroma in this case of importance as regards the development of malignancy?"

Case II. A female patient, aged 30 years, came to the out-patient department in *October, 1954,* with a granulomatous lesion in her nose which was suspected to be a rhinoscleroma. A biopsy was sent to the Pathology Department for verification of the diagnosis and the report issued was as follows (585/54) "This is essentially a rhinoscleroma, but the covering epithelium shows marked hyperplasia, loss of polarity of the cells, hyperchromation and mitosis of the nuclei and early invasion of the deeper tissues (Fig 2). These appearances are an indication of an early malignant change in the covering epithelium."

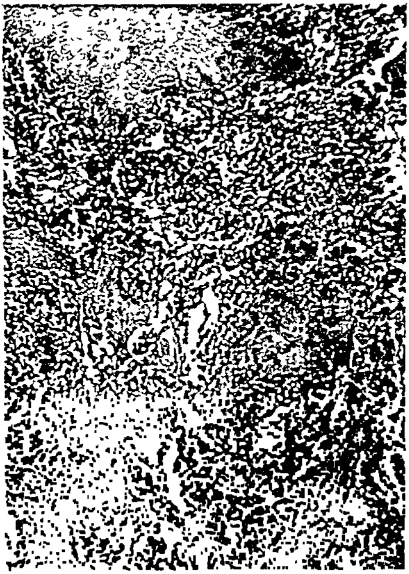

FIG I

A typical rhinoscleroma with numerous Mickulicz cells and some Russell bodies partly covered with stratified squamous epithelium

Discussion

The association of rhinoscleroma and cancer in one lesion at once raises the question of whether there is a relation between the two diseases, i.e. whether rhinoscleroma predisposes to malignancy or the association is only a matter of coincidence. Amongst other workers I have seen a large number of cases of rhinoscleroma but I did not come across or read about cases associated with malignant change as the two instances recorded above. The only known example of a granuloma that may be complicated by malignancy is lupus vulgaris. Rhinoscleroma runs a prolonged course and in the large majority of cases examined histologically there is definite metaplasia of the covering epithelium into stratified squamous and there is also evidence of secondary microbic infection. If we take the analogy of those cases of epidermoid carcinoma which may originate in bronchiectatic cavities, we will find similar conditions in the two cases. In both, the chronic irritation and sepsis produce

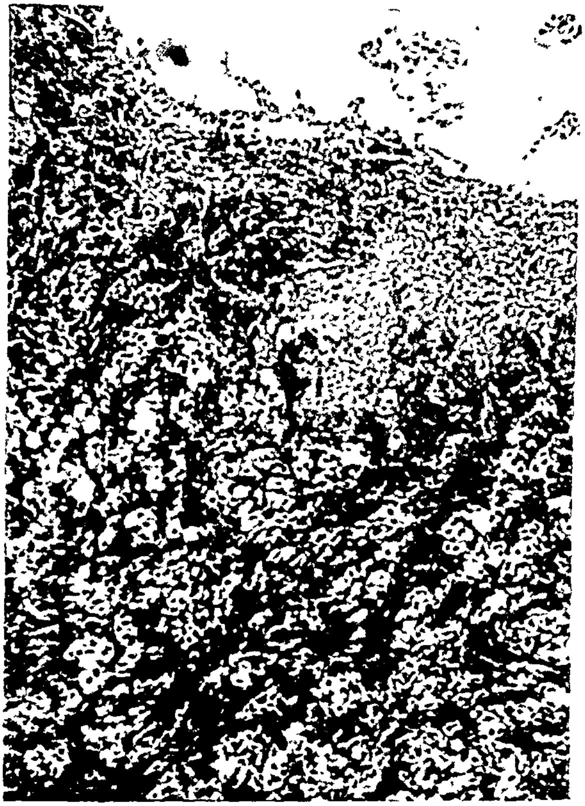

FIG. 2.

This is essentially a rhinoscleroma but the covering epithelium shows marked hyperplasia, loss of polarity of the cells, hyperchromation and mitosis of the nuclei and early invasion of the deeper tissues.

squamous metaplasia of an epithelium which is normally columnar and then in the long run, abnormal and active hyperplasia of this squamous epithelium leads to the development of a squamous cell carcinoma

I have searched the available English literature for cases similar to those recorded above but no such cases were found Therefore malignancy supervening on rhinoscleroma must be regarded as a rare occurrence—though some cases were probably overlooked

Summary

Two cases of rhinoscleroma associated with cancer are recorded To the writer's knowledge they are the first cases showing this association to be recorded in the English literature It is thought that the two lesions are not present together by mere coincidence but that the malignant change has resulted from the long-continued irritation and sepsis due to the presence of rhinoscleroma

TUBERCULOMA OF THE NOSE

By AHMAD EL-HAKEEM (Cairo)

TUBERCULOUS affection of the nose is uncommon. It is considered to be practically always secondary to lung affection. Primary tuberculosis of the nose or sinuses, apart from lupus, does occur but very rare. Only five cases have been recorded by Havens, StClair Thomson mentions two cases.

The reason for the rarity of nasal tuberculosis in comparison to the respiratory tract is due to the structure of the nasal mucosa, the respiratory movement of the cilia, and the bactericidal nature of the secretion.

Location: Most cases recorded affected the nasal septum. The lateral wall is seldom implicated.

Types: *either nodular or ulcerative.*

Lupus may affect the nose at the region of the mucocutaneous junction of the nasal septum with nodules on the face.

I have not come across the literature of nasal tuberculoma combined with traces of healed childhood caseating adenitis.

This rather interesting and rare case of tuberculoma of the nose is worth recording.

FIG I
Tumour after removal.

Case Report

Female, 20 years, pregnant, was admitted to Demerdash Hospital.

Complaint: unilateral partial obstruction of the left side of the nose, occasional bleeding. No discharge and no pain.

Duration: one year. Onset gradual

Past-History: no history of hæmoptysis or losing weight, and no history of admission to a sanatorium.

Tonsillectomy, in childhood
No cough
Suppurating glands of neck incised at the age of 10
General condition good Pulse, 80 Temperature 36 8 °C

Examination of the Nose Purplish red nodular mass limited to the lateral wall of the left nasal fossa, the exact site and the origin was not very clear The mass was rather firm in consistency, not ulcerated and does not bleed on touching The size of the mass was about 3 by 2 cm (Fig 1) There was not any connection between this nodular mass and the septum Right side of nose normal X rays of sinuses no abnormality Post nasal space, normal

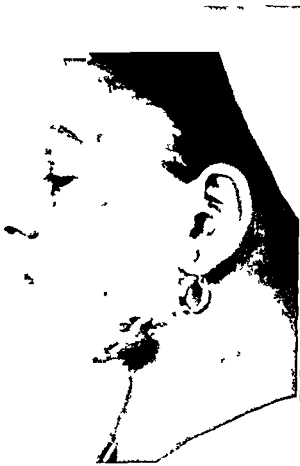

FIG 2
Scar in neck

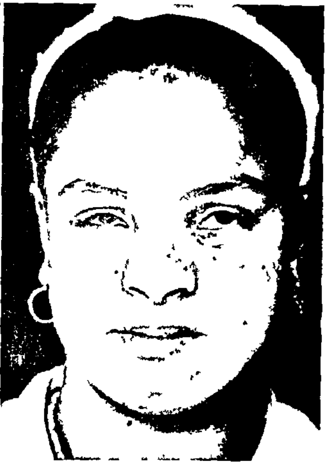

FIG 3
Scar of left lower eye lid

Larynx Normal

Neck Several scars related to adenitis—some at angle of left jaw, and one scar at posterior left sternomastoid group (Fig 2)

Face Scar medial to the inner canthus, with fibrosis pulling the canthus medially and downwards—resulting from an old dacrocystitis (Fig 3)

Another scar below left lower lid margin

Investigations
(1) Wasserman test Negative
(2) X rays of chest No abnormality
(3) Sputum for T B Negative
(4) Smear for T B Negative
(5) Tuberculin test Positive

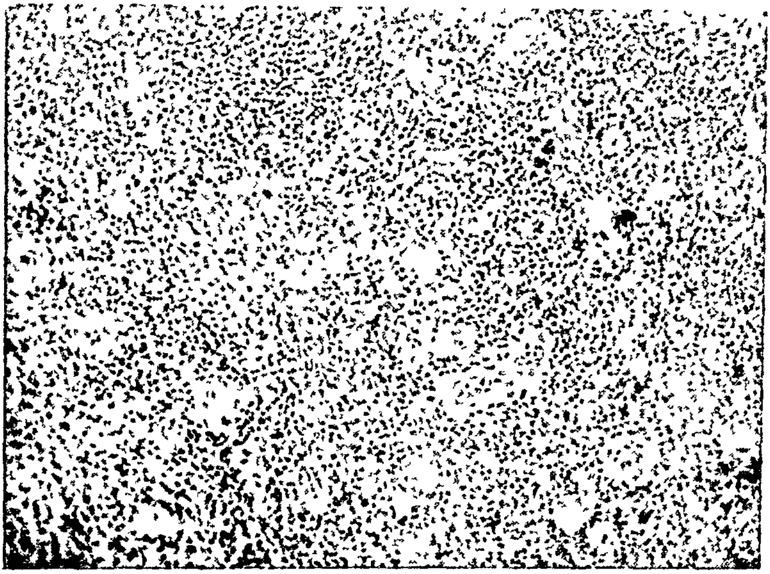

FIG 4
Section of the tuberculoma.

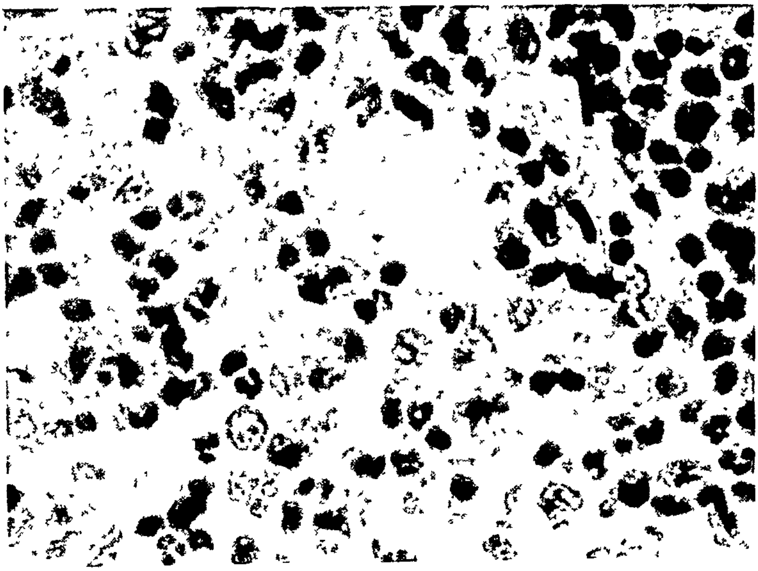

FIG 5
Higher magnification of the tuberculoma

(6) Biopsy Microscopic examination reveals the presence of several tubercles with central Langhan giant cells surrounded by epithelial cells—secondary pyogenic infection is present (Figs 4 and 5) Bacteriological examination Negative for acid fast bacilli

Result Histological picture is consistent with that of *tuberculosis*

(7) Sedimentation rate 1st hour, 8 mm., 2nd hour, 24 mm

(8) Serum protein fractions

Serum total proteins, 7 2 gm per cent
Serum albumin, 4 9 gm per cent
Serum globulins, 2 3 gm per cent
Fractionation of globulins
A globulin, 0·5 gm per cent
B globulin, 0·8 gm per cent
P globulin, 1·0 gm per cent

Treatment Dihydrostreptomycin injections one gramme daily for fifteen days This was followed by operation under local anæsthesia The mass as a whole was removed and the site thoroughly cauterized by galvano-cautery The patient was seen after delivery 90 days later, with a free and normal nose She is under regular observation In these cases removal is usually not followed by recurrence

Comment

This case presents the following features

1 A unilateral nasal obstruction by an unusually large nodular mass, purple in colour, in a female of 20 years

2 The mass was found during operation to be arising from the lower edge of the middle concha, occupying the interconchal space, and partially covering the inferior concha Histological examination of this mass proved to be of tuberculous nature

3 No primary focus was found in the lungs Only scars of ? tuberculous lesions in different parts of face and neck

4 The clinical interest in this case is that the lesion in the nose is not secondary to lung affection, and is, therefore, most probably primary

5 The facial and neck manifestations are on the same side of the face as the mass, the other side is free

6 In spite of the fact that the case is a tuberculoma, yet no change in the serum protein fractions, or in the erythrocyte sedimentation rate is present

I am indebted to Professor Aydarous and to Dr Awny for their kind collaboration in the pathological and laboratory reports

I am also thankful to Elsayed/Abdel-Aziz Noor for the photos and the microphotos

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CLINICAL NOTE

THE EMBRYOLOGICAL ORIGIN OF MIDLINE NASOPHARYNGEAL CYSTS

By J. KEOHANE (Stettler, Alberta)

MILLS (1955) described a midline cystic tumour of the nasopharynx which he stated was similar in structure to the lateral branchiogenic nasopharyngeal cysts described by Taylor and Burwell (1954).

The cysts described by the latter writers are also similar in structure to the better known branchiogenic cysts of the neck. An excellent description of these has been written by Montreuil (1955).

Mills is of the opinion that the cyst described by him probably originated in remnants of Rathke's pouch. It is my opinion that this is an illogical deduction and that the cyst is far more likely to have arisen in remnants of Seessel's pouch. This structure may persist in the adult nasopharynx as the pharyngeal bursa and indeed Mills's description of the cranial extension of the cyst he described, after removal of its dome, fits in with the classical anatomical description of this structure which is said to be occasionally present in the adult and constantly present in the child (Cunningham, 1943). Rathke's pouch or the hypophyseal duct originates in the roof of the oral pit cephalic to the buccopharyngeal membrane which separates the oral pit from the fore-gut in the embryo. On the other hand the pharyngeal pouches develop from the walls of the fore-gut caudal to the buccopharyngeal membrane. The median posterior pouch is Seessel's pouch (Cunningham, 1943a). The former is of ectodermal, the latter of entodermal origin.

Rathke's pouch tumours are usually supra-sellar but may originate and be confined in the sella. They are usually epidermoid carcinomata but may be basal cell cancers of the adamantinomatous type. Cystic degeneration is common (Boyd, 1947). Hyman (1947) describes these cysts as varying in consistency from small solid growths to large multilocular cysts.

I have never seen a case of such a tumour of the nasopharynx arising as a primary condition described but I suppose that such is at least possible. That matter is, however, irrelevant in this discussion. The structure of branchiogenic cysts has been described in detail by the first mentioned four writers and repetition here would serve no purpose. It is, however, radically different to that of Rathke's pouch neoplasms.

Pharyngeal cysts usually arise from either residual adenoid tissue or the pharyngeal bursa (Jackson and Jackson, 1947). Further, since cysts of remnants of Seessel's pouch are really branchiogenic cysts one would expect their walls to be similar histologically to the other cysts of branchiogenic origin (lateral pharyngeal and neck) A midline cyst of the nasopharynx of such a structure is in my opinion almost certain to have arisen from Seessel's pouch, not from Rathke's pouch which is a completely different structure embryologically and gives rise to a completely different kind of tumour, both in structure and location

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SOCIETIES' PROCEEDINGS

ROYAL SOCIETY OF MEDICINE—SECTIONS OF LARYNGOLOGY AND OTOLOGY

COMBINED SUMMER MEETING AT THE UNIVERSITY OF BRISTOL

June 27th, 28th and 29th, 1957

LARYNGOLOGY SECTION

President: J. ANGELL JAMES, M.D., F.R.C.S.

THE opening papers by Miles Taylor, J. M. Yoffey, R. W. Raven, M. Lederman, R. G. Macbeth and Seymour Jones, will be found on pp. 365-408.

DR. A. MACKENZIE ROSS: I thank Dr. Taylor for his most interesting paper. I would like to ask him how it is that in the treatment of Hay Fever, some cases respond admirably to Zinc Ionization while other similar cases do not respond. I wonder if Dr. Taylor would also study the subject—Nasal Polypi—why are they produced and why do they recur after apparent complete removal? I personally have also observed that in cases of Nasal Catarrh and Allergic Rhinitis, the administration of Calcium has a beneficial effect and lessens the catarrh. This would appear to confirm that there is a chemical abnormality in the tissues in conditions of Nasal Allergy.

DR. MILES TAYLOR: I regret that my work is not sufficiently advanced to give a detailed reply to your query.

I would like to express my thanks to the Society for the kind way in which they have received my communication.

MR. MACBETH asked Professor Yoffey: If the tonsils had been shown to have afferent lymphatic vessels.

In reply to Mr. R. G. Macbeth, PROFESSOR YOFFEY said that while we had not made any special study of tonsillar lymphatics, we had never, in the type of experiments which we performed, seen evidence of dye entering the interior of the tonsil, even though the peri-tonsillar network was well marked and heavily stained. He pointed out, too, that the structure of the tonsil is such that it is not placed in the course of a lymph stream, and does not possess the lymph sinuses which seem to be characteristic of lymphoid tissue which is receiving afferent lymphatic vessels.

MR. R. C. TUDWAY: The treatment of carcinoma in the pharynx and larynx by deep X-ray therapy has always been restricted because the tissues in that area are sensitive and react easily to radiation, while the growth is fairly resistant and requires quite a heavy dose to give any chance of cure. In the case of carcinoma of the larynx X-ray treatment has had a little success, but in the case of post-cricoid carcinoma the difficulties mentioned are greater and our efforts with deep X-rays have been consistently ineffective A better quality of radiation, such as that from the tele-radium unit Dr Lederman has used, or the radioactive cobalt version of this machine, does however have greater success as Dr Lederman has shown

I find myself in complete agreement with what Dr Lederman has said except possibly for one minor point I think there is some advantage in the larger and heavier machines such as the large tele cobalt units or linear accelerators This is I think, a minor advantage arising from the fact that the skin reaction when these larger machines are used, is a little less and the beams can be directed with slightly greater precision These slight advantages would certainly not justify the installation of this large machinery merely for the treatment of post-cricoid lesions, but when this kind of equipment is available I feel sure that the larger cobalt units or 4 million volt linear accelerators are the best forms of radiotherapy that we have at present

Our own experience at Bristol has been just as depressing as that of other centres and is based mainly on the use of orthodox deep X-rays My colleague, Dr Curwen, will be telling you a few details We have, however, in the last two years been using a full sized cobalt beam unit for these cases My first impression is that the patients are doing a little better with this treatment, because the radiation reactions are very much less and a more effective dose may be given We shall have to wait of course, to see if these first impressions are correct and whether we can salvage a few more cases by the use of the higher quality radiation

MR SYDNEY CURWEN The treatment of post-cricoid carcinoma with radiotherapy has hitherto proved unsatisfactory Using conventional, that is to say, deep X-ray therapy in the 250 K V range, very few patients survive more than a year, and only two patients out of some thirty-five cases reviewed over eighteen years at Bristol, survived more than five years The patients treated with the tele cobalt unit have shown a slightly greater promise of success, but as this unit has only been in operation for two years it is a little early as yet to assess the results I agree with Dr Lederman that a combined attack of radiotherapy of the higher energy type followed by surgery is the method of choice I do feel, however, that this procedure must be carefully planned and adhered to even if the immediate results of therapy are successful surgery should follow in every case

MR M FAIRMAN I would like to present a report upon the fate of all cases of post cricoid carcinoma of which record can be found, referred to the E N T Department of the Bristol Royal Hospitals during the past ten years

The treatment of these cases was considered with a definite bias in favour of surgical treatment, and, therefore, the more promising cases were selected for operation and the more advanced cases and those with poorly differentiated growths, were referred for radiotherapy For this reason the end results of treatment as reported in this communication may differ from those reported by my colleague of the radiotherapy department

The total number of cases I have to report is 49 15 male and 34 female, in all the diagnosis has been confirmed histologically The survival rate has been calculated from the date of diagnosis Of these one only, i e 2 per cent , has survived 5 years, but 5 cases (one still undergoing treatment) are still alive today.

It is pointless to talk in terms of "5 year cure" in regard to these figures, but considered in terms of 2-year survival, the results of surgical treatment alone or combined with radiotherapy, with 8 cases surviving out of a total of 23, show a marked advantage over the results of treatment by radiotherapy alone, with 1 survival out of 20.

The choice of cases for surgical treatment has varied considerably with individual surgeons. I personally, have confined my selection to cases of very limited extent, with no glandular metastases and histologically well differentiated. The surgical procedure adopted has varied from (1) laryngo-pharyngo-œsophagectomy with two-stage repair by means of full thickness skin flaps from the neck; (2) laryngo-pharyngo-œsophagectomy with one-stage repair by means of a split-skin graft supported on a plastic tube; (3) pharyngo-œsophagectomy with preservation of the larynx together with two or one-stage closure as above. In a number of cases dissection of the cervical glands has also been performed.

The cases treated by combined surgery and radiotherapy have been either cases of failed surgery or failed radiotherapy, or surgery for the excision of stricture following radiotherapy. The combination of treatment has been largely fortuitous rather than by intent.

All the cases complained of some discomfort in the throat and most of difficulty in swallowing. Two cases presented as thyroid tumours and in one the thyroid was explored. Three cases were diagnosed on a second endoscopy a few months after an original negative endoscopy. Search of the records has revealed one case, treated by radiotherapy alone, which survived 17 years and nine months, and then died of a recurrence.

Cases of Proven Post-Cricoid Carcinoma Referred to the E.N.T. Department of the Bristol Royal Hospital Since January 1st, 1947. To Show Survival to Nearest Six Months

	Total	6 mo.	1 yr.	18 mo.	2 yr.	2½ yr.	3 yr.	3½ yr.	4 yr.	4½ yr.	5 yr.	
All cases	M. F. 15 34 49	32	20	11	9	8	6	4	3	3	1	5 cases still alive, 1 under treatment.
No treatment	6	2	1	1	0	0	0	0	0	0	0	
Radiotherapy alone	20	9	4	1	1	0	0	0	0	0	0	
Surgery alone	12	10	7	4	4	4	2	1	1	1	0	3 still alive after 1 year 3 months, 2 years 7 months, 4 years 7 months.
Surgery with radiotherapy	11	11	8	5	4	4	4	3	2	2	1	1 still alive after 7 years 6 months.

Mr. K. G. Malcomson: Of the 227 cases of carcinoma of the hypopharynx seen in the E.N.T. Department in the Cardiff Royal Infirmary in the past 25 years, 75 were treated by partial or total pharyngolaryngectomy.

In the 20 year period up to June, 1952, 43 patients underwent pharyngolaryngectomy Thirteen of these patients died within a month of operation (12 within the first post operative week), i e 32 per cent of the total died during the immediate post operative period

The deaths during the first month are largely ascribable to the effects of operation, and those that survive this period are more likely to succumb to recurrence of the disease Thus, of the group of 30 patients surviving the immediate post-operative period, 18 (60 per cent) survived 1 year, 10 (33 per cent) survived 2 years, 9 (30 per cent) survived 3 years, 8 (27 per cent) survived 4 years, and 6 (20 per cent) survived 5 years or more, or nearly 14 per cent of the original group of 43 patients The longest survival period without recurrence is 23 years

If the whole group of 75 patients is taken, there were 24 deaths (32 per cent) during the immediate post operative period Of the 51 patients surviving this period, 31 (62 per cent) survived 1 year 13 (26 per cent) survived 2 years, 10 (20 per cent) survived 3 years, 8 (16 per cent) survived 4 years, and 6 (12 per cent) survived 5 years or more

The use of antibiotics in the management of patients subjected to pharyngo laryngectomy would be expected to be of most value in the immediate post-operative period, i e the first month The advent of the general use of anti biotics about 12½ years ago gives a natural division of the 25 year period into equal halves Up to the end of 1944, Mr Owen had carried out pharyngolaryngectomy on 15 patients, 6 of whom died within a month, a mortality of 40 per cent Of the 60 pharyngolaryngectomies performed by Mr Owen since then, there were 18 deaths during this initial period, a mortality of 30 per cent The reduction in mortality by 10 per cent is not very significant, undoubtedly nursing skill remains of fundamental importance during the critical first month

Mr Lewis *suggested that in the two-stage operation*, closure could be effected in three to four weeks, rather than waiting for two months, as Mr Raven recommended

Mr R W Raven in reply to Mr R S Lewis I leave a period of two months between the two operations to allow the tissues to settle down and the new skin flap to acquire an adequate blood supply

If pre operative radiation is given serious surgical problems are created which tax the resources of the surgeon to the utmost Having in mind the great suffering of these patients we must proceed carefully with surgical treatment for which patients must be selected These patients are amongst the happiest I have and I feel sure that with earlier diagnosis the results will improve

Professor F C Ormerod said that he considered the outlook for patients with post cricoid cancer to be very much better, than even a few years ago The results of one stage excision described by Shaw and himself in March in the *Journal of Laryngology and Otology* showed seven survivals for one year or more out of seventeen These figures dated to August, 1956, but the seven patients were still well almost a year later, and they had all been fairly advanced cases Dr Lederman had reported seven out of seventeen three-year survivals without recurrence in early cases and it might be that there was a place for radio therapy in demonstrably early cases, but advanced cases should be treated by primary surgical excision There was no doubt that the post-operative period was much shorter and recovery more complete if the patients had not been previously irradiated.

He was very sorry to hear one member refer to the misery and wretchedness of these patients after operation, and suggest that it was kinder to leave them alone. This was quite untrue. The patients were relieved of a septic fungating cancer, were up and about the day after operation, were swallowing in two weeks and many went home in three weeks. They were comfortable and happy, were able to lead useful lives and were very grateful for what had been done to them. There was very little in the neck that it was not possible to remove, and as it was now quite feasible to construct a new gullet immediately, there seemed to be no excuse for not carrying out this treatment.

MR. GEORGE SEED reported from Leeds:

Number of Patients seen at the Leeds General Infirmary (1946-1955)		58
Sex Incidence:		
Males	3	
Females	55	
Age Incidence		
30-40 years	2	
41-50 years	12	
51-60 years	15	
61-70 years	16	
71-80 years	12	
81-90 years	1	
Treatment:		
No treatment, too advanced	15	
Patient refused treatment	1	
Palliative Deep X-ray	11	
Deep X-ray Treatment	20	
Radium Beam	9	
Radium Collar with bougie	2	
Results:		
Six year cure	1	
Five year cure	1	
Died	56	

Of the 56 patients who died four patients remained well for 15 months, 18 months, 18 months and 2½ years respectively.

MR. A. J. MOFFETT: The Cancer Follow-up Department of the United Birmingham Hospitals has very kindly given me an extract of 139 cases which came under the care of the Hospitals during a period of 17 years, prior to 1955, and were subjected to various forms of treatment, radical or palliative, surgical or radio-therapeutic.

It seems apposite that this discussion is on the Management, rather than the treatment of post-cricoid carcinoma, for of this number (139), there were no survivors for the five-year period, and only 9 for more than one year. One case of this series survived 4½ years, only to die of a radiation myelitis of the cervical chord, and I know of another, outside the range of this series, which has survived to develop a quadriplegia from the same cause.

My contribution, to this unhappy catalogue was an attempt in 1950 to improve on these results by radical surgery, comprises 9 cases operated on during the next 3 years.

The first 3 cases were two-stage operations. One of these developed a recurrence before the reconstruction phase could be undertaken, and presented a picture of such indescribable misery, that I felt some form of one-stage operation must be devised which would obviate this awful happening

The next 6 were one-stage operations of various types, and of the total of 9 cases, 6 had either uni-lateral, or bi-lateral block dissection of the glands of the neck I do not think that this procedure makes the slightest bit of difference

None of my cases had a gastrostomy, nor was it necessary I felt that with 3 unnatural orifices in the neck, the patient should be spared yet another in the abdomen

Of my one stage cases, one lived and returned to run his business for 2½ years—apparently a surgical triumph—only to die of a histologically unrelated carcinoma of the bronchus, to which laryngectomized patients seem peculiarly susceptible

This last case made me so disheartened that at that time, I felt that I would never again operate on one of these cases, and felt that the hope of cure must lie within the field of radiotherapy I think that I can say with all fairness, that within my experience that hope has not yet been fulfilled

If I do make a further attempt, it will be on these lines The preliminary diagnosis would be made by inspection only I suspect that a biopsy confirms, not only the diagnosis, but the patient's fate

The growth and the gland area would be irradiated with a dose sufficient to halt the tumour, and discourage dissemination, but not sufficient to devitalize the tissues and make subsequent surgery hazardous This would be followed by a two stage operation without block dissection

We all must feel that the published figures on this condition can give us little cause for satisfaction, but we tend to publish our successes, and keep our failures to ourselves Therefore, it is not the published, but the un published figures which will give the truer picture

It needs very little imagination on the part of those who have had the care and treatment of these cases, to see in the wards and departments, surgical and radiotherapeutic, throughout the country, an aggregate of human misery, at which one is appalled

I have great doubt in my mind whether it is justifiable to subject 5, 10, or 20, or whatever the number is, of patients to the misery and pain of surgery, or radiotherapy, prolonging their lives for a few weeks or months of unrelenting misery, in the hope of producing one survivor At present I feel that the kindest thing to do in many of these cases is probably—nothing

An extract of the survival rates of 139 cases of post cricoid carcinoma under the care of the United Birmingham Hospitals

1936 1953

Females	100	Males 39
Untreated	4 cases Because of age	
Remainder	135 cases All types of treatment	
Survived	1 year or more	4
Survived	2 years or more	4
Survived	3 years or more	Nil
Survived	4 years 9 months	1 with quadriplegia

No 5 year survivors

GENERAL NOTES

THE Editors are anxious to improve the clinical aspect of the Journal and would welcome more reports of interesting cases. These clinical records need not be at all elaborate and it would be unnecessary to go minutely into the ætiology or the literature of the subject. Senior Registrars have unique opportunities for seeing a large variety of cases, many with unusual features. It is in many ways regrettable that these should go unrecorded. It is hoped that all those in charge of clinics will help us in this matter and see that such cases are not overlooked.

OTOLARYNGOLOGY LECTURES 1957-58

THE following lectures, arranged jointly by the Royal College of Surgeons of England and the Institute of Laryngology and Otology, will be delivered in the Lecture Hall of the College, at 5.30 p.m.

1958
July 3rd. Mr. Norman C. Tanner. Haemorrhage from the Œsophagus.

INTERNATIONAL CONGRESS ON THE MODERN EDUCATIONAL TREATMENT OF DEAFNESS

AN International Congress on the Modern Educational Treatment of Deafness will be held at the University of Manchester from July 15th-23rd, 1958.

The Congress is being organized with the approval and support of the Council of the University, with financial help from the Ministry of Education, the National College of Teachers of the Deaf, and the Rathbone Trustees, and with the co-operation of the British Council and the Alexander Graham Bell Association for the Deaf, Inc., Washington, D.C., U.S.A.

The Congress is being planned to bring together educators, otolaryngologists, pædiatricians, medical officers, audiologists and educational psychologists, to consider and discuss the significance, to the future development of their work, of the results of modern research on problems of deafness in childhood.

Arrangements are being made to offer accommodation to members of the Congress in University Halls of Residence. The cost of this type of accommodation is expected to be about £1 2s. per day, including bed, breakfast and dinner. The University Refectory will be available for meals during each day.

Forms of application for membership of the Congress will be supplied upon request, to Professor A. W. G. Ewing, Department of Education of the Deaf, the University, Manchester 13, England.

General Notes

THE BELGIAN SOCIETY OF OTOLOGY, RHINOLOGY AND LARYNGOLOGY

From 2nd to 7th June, 1958, an Extraordinary Congress, under the Patronage of the Commissariat General of the Belgian Government will be held at Brussels, at the Universal and International Exhibition, Brussels, 1958 President, Prof J van den Branden (Brussels)

SCIENTIFIC PROGRAM

(1) Cancer of Larynx First European meeting of the International Commission for the Study of the Cancer of the Larynx, (2) Functional Surgery of the Ear, (3) Meeting of the Societe de broncho œsophagologie de Langue Française, (4) Meeting of the International College of Experimental Phonology, (5) Festival of scientific film in E N T field

Every participant in the Congress will be granted an A1 priority concerning accommodation and free access to the exhibition from 1st to 8th June

The social program and all other informations will be communicated later

Inscription fees Effective members Fb 500, Adherent members Fb 250

For any information, kindly apply to the General Secretary, Dr Henschel, C E L, 19 Rue de Fetinne Liege, Belgium

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FACULTÉ DE MÉDECINE DE PARIS

Chaire de Clinique Oto Rhino Laryngologique Professeur A Aubin Cours de Technique Chirurgicale—La Chirurgie de la Surdite Lundi, 9 au Samedi, 14 Juin, 1958

Apply Pavillon Isambert, Hôpital Lariboisiere, 2 Rue Ambroise-Pare, Paris

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ROYAL SOCIETY OF MEDICINE

SECTIONS OF LARYNGOLOGY AND OTOLOGY

Summer Meeting in Edinburgh, June 27th, 28th 1958

Friday, June 27th

9 30 a m Scientific Meeting Royal College of Surgeons of Edinburgh, Nicholson Street *Discussion* "Acoustic Neuroma" Professor Norman Dott Dr C S Hallpike Dr D Dix

1 p m Luncheon for Members and Guests Carlton Hotel, North Bridge Price 15s inclusive of wines and gratuities

Afternoon Garden Party by invitation of the Presidents

7 15 for 7 45 p m Dinner in The Royal College of Surgeons of Edinburgh Tickets Two guineas each inclusive of wines

Saturday, June 28th.

9.30 a.m. Scientific Meeting: Royal College of Surgeons of Edinburgh. *Discussion:* "The dysphonias and the scope of speech therapy and their treatment". (Illustrated by recordings). Mr. Maxwell P. Ellis. Mr. L. Willmore.

12.30 p.m. Meeting ends.

For details of accommodation write to the Secretaries of the Sections.

Registration Fee of 10s. will be made for each Member to cover overhead expenses.

Members wishing to attend the Meeting should apply *as soon as possible.* Bookings at hotels must be made through the Royal Society of Medicine and *not* directly with the hotels. Accommodation is limited and early application should be made.

Applications should be accompanied by a cheque *made payable to Dr. J. F. Birrell* to cover the registration fee, lunch and Dinner tickets, but *not* for hotel accommodation.

N. L. CRABTREE, Honorary Secretary, Section of Laryngology.
LIONEL TAYLOR, Honorary Secretary, Section of Otology.

BRITISH ASSOCIATION OF OTOLARYNGOLOGISTS

MINUTES OF THE ANNUAL GENERAL MEETING HELD AT THE ROYAL SOCIETY OF MEDICINE, 1 WIMPOLE STREET, LONDON, W.1, ON FRIDAY, DECEMBER 6TH, 1957 at 4.30 p.m.

Present: Mr. I. Simson Hall (President) in the Chair and 41 members.

1. *Minutes* of the meeting held on December 7th, 1956, were read and signed as correct.

2. *Election of Members.* The following were elected members on the nomination of the Council:

Henry Douglas Fairman, Bristol.
James Littlejohn, London.
William McOnie Macgregor, Gravesend.
Kenneth Greer Malcomson, Bristol.
Raymond Hatfield Welch, London.

3. *Report of Council for 1956-7.* The report of Council for 1956-7, which had been circulated, was discussed and approved.

4. *Report of the Honorary Treasurer and the Balance Sheet.* The audited accounts for *1956-7*, which showed a surplus on the year's working of £30. 5s. 8d., were presented by the Honorary Treasurer and approved.

5. *Election of Officers.* The following were elected Officers for 1957-8:

President:	Mr. C. Gill-Carey.
Immediate Past President:	Mr. I. Simson Hall.
Vice-President:	Mr. R. R. Simpson.
Honorary Treasurer:	Mr. J. C. Hogg.
Honorary Secretary:	Mr. Myles L. Formby.

6 *Election of Members of Council* The election of the following members of Council to the six vacancies was confirmed

London	Mr R J Cann
	Mr I G Robin
Northern England	Mr J H Otty, Bradford
Southern England	Mr R L Flett, Derby
	Mr Gavin Livingstone, Oxford
Scotland	Dr J Fulton Christie, Glasgow

7 *Auditors* Messrs Howard Howes & Co , were re elected Auditors for the ensuing year

EXTRACTS FROM THE REPORT OF COUNCIL

Poliomyelitis Inoculation and Tonsillectomy

The Ministry of Health has now stated that they do not lay down that any special interval should elapse between poliomyelitis inoculation and the removal of tonsils and adenoids This will, in effect, cancel the notice circulated to members in June 1956 in which a six weeks' interval was recommended

World Medical Association, British Supporting Group

The Association has been invited to join the British Supporting Group of the World Medical Association It has, however, been agreed that at the present time no useful purpose would be served by the Association joining this Group as a corporate body and that it should be left to members to join as "ordinary members", if they so desire

Ruedi Trust

No further grants have been made from the Ruedi Trust Fund and the balance of £19 1s 3d remains in the bank

The Council has agreed that the Fund shall now be closed and that the balance shall be offered to any young otologist undertaking post graduate study in Switzerland

Royal College of Surgeons of England

The Royal College of Surgeons has asked the Association to subscribe to certain periodicals on behalf of the Library The Council has agreed to subscribe to journals approved by the Library Committee up to a sum not exceeding four guineas per annum

Diary of Ear, Nose and Throat Lectures

The Association is now keeping a diary of all ear, nose and throat lectures and/or demonstrations of public interest held in London and also the dates of meetings of the Visiting Associations

General Notes

ROYAL COLLEGE OF SURGEONS OF ENGLAND

ORDINARY COUNCIL, DECEMBER 12TH, 1957

REPORT OF THE COURT OF EXAMINERS

THE Court of Examiners reports to the Council that at the recent Final Examination for the Fellowship, 27 candidates presented themselves for the Examination in Otolaryngology, 5 of whom acquitted themselves satisfactorily.

The following are the names of the 5 candidates who are therefore entitled, subject to the approval of the Council, to the Diploma of Fellow:—

Name	Qualification	Address
Singh, Daryao	M.B., B.S. Punjab	c/o Military Adviser to High Commissioner for India, 76 South Audley Street, W.1.
Sinha, Achyutananda	M.B., B.S. Patna	"Krishna Bhawan", Frazer Road, Patna-1 (Bihar), India.
Holborow, Christopher Adrian	M.B., B.Ch. Camb.	4 Weston Rise, W.C.1.
Dunn, John Talbot	M.B., B.S. Sydney	3 Curraghheena Road, Mosman, New South Wales, Australia.
Griffith, Adrian Nicholas	M.B., B.Ch. Camb.	Bee House, Bolney, Haywards Heath, Sussex.

The Journal of
Laryngology and Otology

(*Founded in* 1887 *by* Morell Mackenzie *and* Norris Wolfenden)

June 1958

HYPERNASALITY (RHINOLALIA APERTA) FOLLOWING TONSIL AND ADENOID REMOVAL

By A G GIBB (Dundee)

I FIRST became interested in the subject of hypernasality four years ago, when a boy of 6 years (Case I) whose tonsils and adenoids I had removed a few weeks previously, was brought back to see me on account of "bad speech" At my pre operative examination, I noted very large tonsils and a moderate amount of acutely inflamed adenoids, which I took to be the cause of his recent acute otitis media My case notes gave no further information about the throat I advised removal of tonsils and adenoids The parents assured me that the boy's speech was normal prior to operation The speech now resembled that of a patient with a severe cleft palate On examination, I found a normal tonsil and adenoid result with no damage to faucial pillars, no distortion or scarring of palate, and a normal posterior pharyngeal wall A double uvula was present, and this was associated with an excessively short palate and a wide gap between the soft palate and the posterior pharyngeal wall The boy was unable to effect closure of the nasopharynx when talking and nasal escape was severe Speech therapy and blowing exercises were commenced immediately, and within 2-3 months there was a marked improvement in speech Despite this, the boy's father was dissatisfied and legal action was only averted by a tactful general practitioner The boy was referred to a plastic surgeon for opinion regarding operative narrowing of the palato-pharyngeal sphincter

The surgeon thought it unwise to operate in view of the parent's attitude and advised further speech therapy. This was continued with improvement. I feel that this boy, although much improved, cannot achieve normal speech without additional operative treatment, for there is still well marked nasal escape despite good palate movement.

The stimulus derived from this case led me to investigate all cases of hypernasality requiring treatment following tonsillectomy and adenoidectomy in the Eastern and part of the South-Eastern Regions of Scotland. The area under investigation has an average population of 620,000. During the period 1950-57 27,734 tonsil and adenoid operations were performed in children. 19 children developed hypernasality immediately after the operation. The operations performed on these children were carried out by 11 different surgeons, seven of whom were consultants with considerable experience. It will thus be readily appreciated that any individual surgeon has an extremely limited experience of this complication. *The figures suggest that open nasal speech develops in approximately* 1 : 1,450 tonsil and adenoid operations. The incidence, however, is probably considerably higher than this, for children with slight or transient defects are unlikely to report back for treatment. In addition, I was obliged to exclude 12 other cases of hypernasality from this series for the following reasons:

(*a*) Lack of definite information regarding the state of speech before operation.

(*b*) The presence of another speech defect which overshadows the hypernasality.

(*c*) Operative damage to palate or pillars of fauces (whether this was considered to be the cause of the hypernasality or not).

This investigation is limited to cases where the tonsils and adenoids have been efficiently removed with no damage to adjacent structures. No blame can be attributed to the operative technique of any of the 11 surgeons concerned.

It is important to realize that, except in rural areas, these patients are seldom referred back to the surgeon concerned but find their way *viâ* the school teacher to the speech therapist, who visits the school. In nearly all cases in this series, the surgeon was unaware that such a condition existed after operation. It is, therefore, very important for the surgeon to keep in close liaison with the speech therapists in his region.

Investigation

The 19 cases of hypernasality were fully investigated. All occurred in children; no cases were encountered following tonsil operations in adults.

History

The parents were questioned regarding the birth and development of the child, any congenital defects being noted Special attention was paid to physical or mental retardation, the intelligence quotient being determined if necessary It was important to get an accurate idea of the child's speech before operation While in a few cases a speech therapist's opinion was available, in the majority of cases one had to rely on the parents' statement If the latter was indefinite or unavailable, the case was not included in the series

Operation Details of the tonsil and adenoid operations were ascertained in all cases, in order to find out if special difficulties were encountered or if post operative complications developed

Examination A careful examination of the child's nose, throat and ears was carried out special care being taken to determine the development, length and mobility of the palate

Hypernasality was assessed by the following methods

(1) Mirror test—The child articulates selected sounds while a mirror, placed under the nose, is watched for misting The sounds normally used are, (*a*) consonants—plosives p, b, t, d, k, g, fricatives f, v, s, z, sh, zh, ch, j, (*b*) vowel sounds These sounds were tested both in isolation and combined in words No misting should occur with any of the selected sounds unless the palatopharyngeal sphincter is incompetent

(2) Reading and conversation tests were carried out to determine the degree of nasal escape in ordinary speech

An independent assessment was made by a speech therapist

X-rays were not taken routinely in this series of cases, but in selected cases careful radiographic study of palate movement and closure may prove valuable in deciding treatment and assessing progress (Figs 1-6)

Treatment The date of commencement, the duration and the type of treatment was noted

Family Investigation All parents were questioned regarding speech defects in other members of the family and the effect of tonsil and adenoid operations in near relatives Where possible I examined the throats of the child's parents and other members of the family In all, 34 relatives were examined

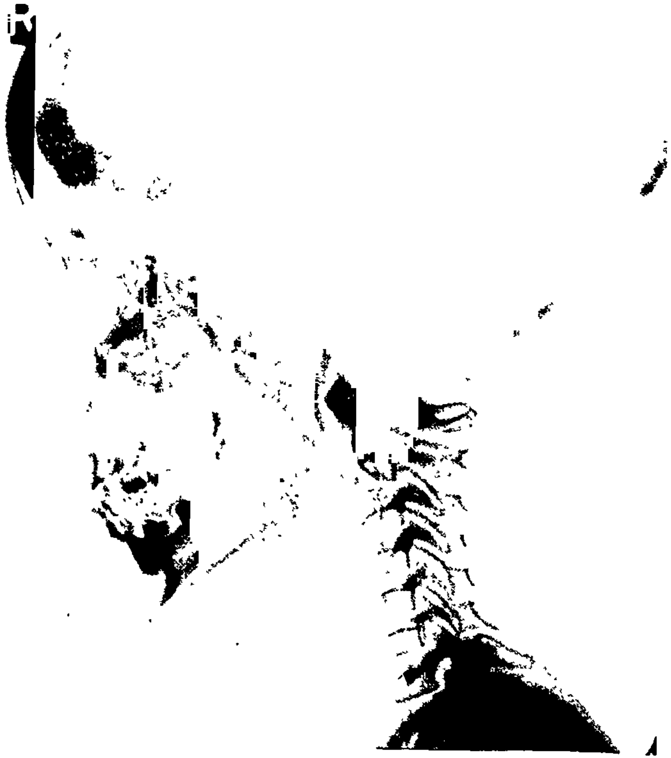


Fig. 1.
Normal palate at rest (fine wire suture marks the free margin of the soft palate).

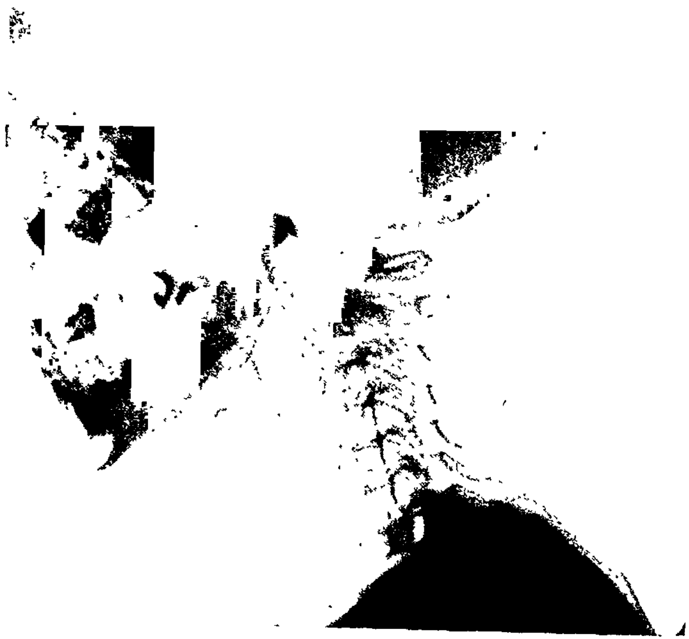


Fig. 2.
The same palate elevated by saying "s . . . s . . . s . . ." showing good sphincter action. (Similar movement can be produced by blowing, sucking and certain other sounds.)

Hypernasality (Rhinolalia Aperta)

Results of Investigation (Table I)

All cases occurred in children between the ages of 3 and 8 years Both sexes were affected equally

Congenital defects included pes cavus, undescended testis and microtia Five cases of retarded mental development occurred in the series In addition, one child was premature and suffered from icterus neonatorum All cases (except Case VII) showed some degree of palate shortening This varied from a slight insufficiency to a gross shortening where the palate was separated from the posterior pharyngeal wall by a gap of 2-3 cms In five cases this was associated with a cleft uvula

Operation The tonsils were removed by guillotine in 18 cases, in the other case the dissection method was employed In the area under investigation, all but a few tonsil operations are performed by the guillotine method in children As has been stated, there was no damage to the palate, faucial pillars or posterior pharyngeal wall Slight tightness of the soft palate was noted in five cases

Post operative complications These included one case of adenoidal hæmorrhage This was apparently severe and required the insertion of a post-nasal pack

Two children suffered from severe nervous upset after operation this was considered an important factor in the production of their hypernasality

Tests for hypernasality Mirror tests for nasal escape showed gross misting in 10 cases and slight misting in the remainder The sounds most severely affected in the majority of cases were sibilants, "s", "sh", but plosives and vowel sounds were, as a rule, very nasal also

Nasal escape was generally easily detected both in ordinary conversation and in reading

Treatment (*a*) *Speech therapy* was commenced soon after operation in eight cases only A delay in starting treatment in the remaining 11 cases resulted in the hypernasality becoming well-established Treatment of rural cases presented a difficult problem, as regular training and supervision by a speech therapist were impossible The affected children were given initial training at the speech therapy centre and exercises were continued under the parents' supervision and guidance This was seldom satisfactory due to lack of firmness or intelligence on the part of the parents

Following treatment 13 children developed a return of good palate movement, while in the other six cases, movement remained poor

In 11 cases marked speech improvement accompanied the return of palate movement If, however, the palate was very short, nasal escape tended to persist in spite of treatment

TABLE I

Results of Investigation—Case Summary

Case No	Age at operation	Congenital Deformities			Pre-operative Speech	Post-operative Therapy		
		Palate shortening	Uvula	Others		Interval before starting	Duration	Result
1	6	++++	Double	—	—	1 month	Home exercises only	Improved
2	5	++++	Bifid	Mentally retarded	—	1 month	6 years	Still very bad
3	5	++	—	—	Stammer	6 months	Home exercises only	Improved
4	4	++++	—	—	—	3 months	18 months	Still bad
5	5	++++	—	—	—	No treatment	—	—
6	5	+++	—	Undescended testis	—	3 weeks	4 years	Improved
7	5	Nil	—	—	—	1 month	4 years	*Unchanged*
8	7	++++	—	Mentally retarded	Slightly hypernasal	1 month	9 months	Improved
9	4	++++	Bifid	—	"thick"	18 months	5 years (intermittent)	Still very bad

Hypernasality (Rhinolalia Aperta)

10	5	++	Double	—	Lateral S Slightly hypernasal	1 month	18 months	Improved
11	7	++++	—	Microtia Pes Cavus	Slightly hypernasal	6 months	1 year (intermittent)	Unchanged
12	6	+	—	—	thick	1 year	7 years (intermittent)	Improved
13	6	++	—	—	thick	1 year	7 years	Almost cured
14	8	++	—	Mentally retarded I Q 54	—	4 months	5 months *(continuing)*	Improving
15	6	++	—	—	—	1 month	(Recent case)	—
16	4	+	—	Mentally retarded I Q 59	bad hypernasal	2 years	1 month	Unchanged
17	4	++	—	—	—	4 years	3 years +	Much Improved
18	3	+	Bifid	Mentally retarded	bad	5 years +	18 months	Cured
19	7	+	—	—	—	3 weeks	9 months	Almost cured

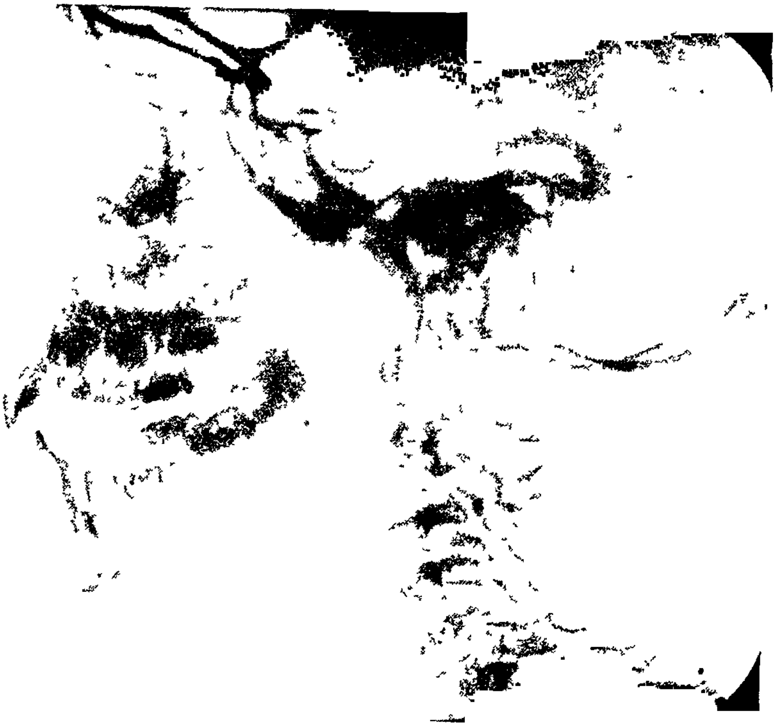


Fig 3
Normal—a shorter palate than in the previous case (tonsils and adenoids have been removed)

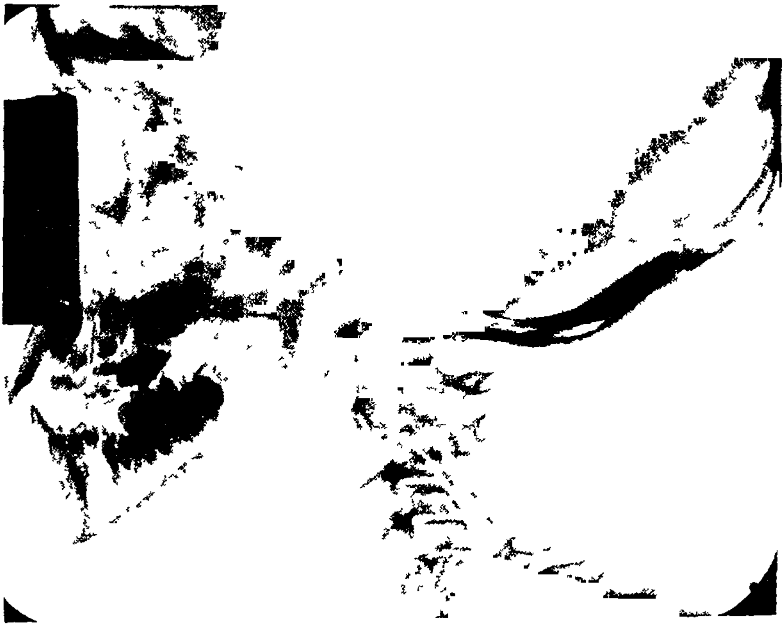


Fig 4
The same patient showing excellent palate movement on saying ‘ s s s and a competent sphincter

(*b*) *Operation.* No children in this series were submitted to operative treatment; this is not surprising when it is realized that only a few cases were referred back to the surgeon concerned

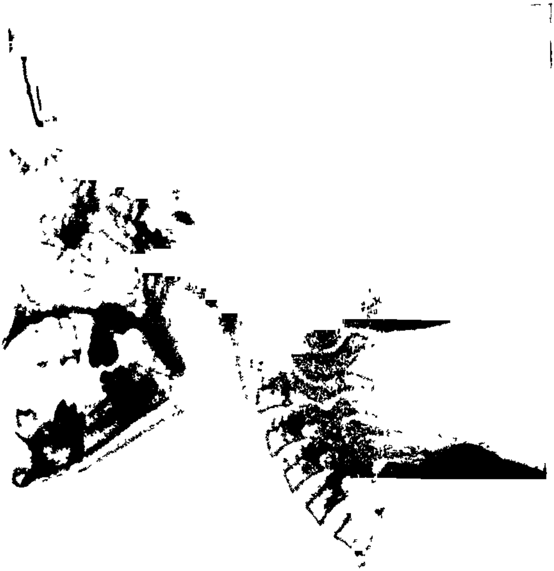


FIG 5
A case of hypernasality (Case II)—at rest (Fine wire suture marks free margin of soft palate) Note the very short palate

Family Investigation In one instance, twin girls both developed hypernasality of equal degree after operation

Open nasal speech also developed in one child's mother and in another child's sister after tonsil and adenoid removal In only two of the 34 relatives examined was there a significant degree of palate shortening. In one of these cases the tonsils and adenoids had been removed without ill-effects

Special Cases

1 (*Case V*)

H S , female, aet 5, underwent a routine examination at the school clinic. Her parents had no complaints about her health, but her mother thought she was dull in hearing at the time. Speech was normal Tonsils (guillotine) and adenoids were removed on 21.9.54, with no complications. Marked hypernasality developed immediately, but no speech therapy was given. This might have been difficult to arrange as the child stayed 20 miles from the nearest centre. When seen on 25.10.55 there was severe

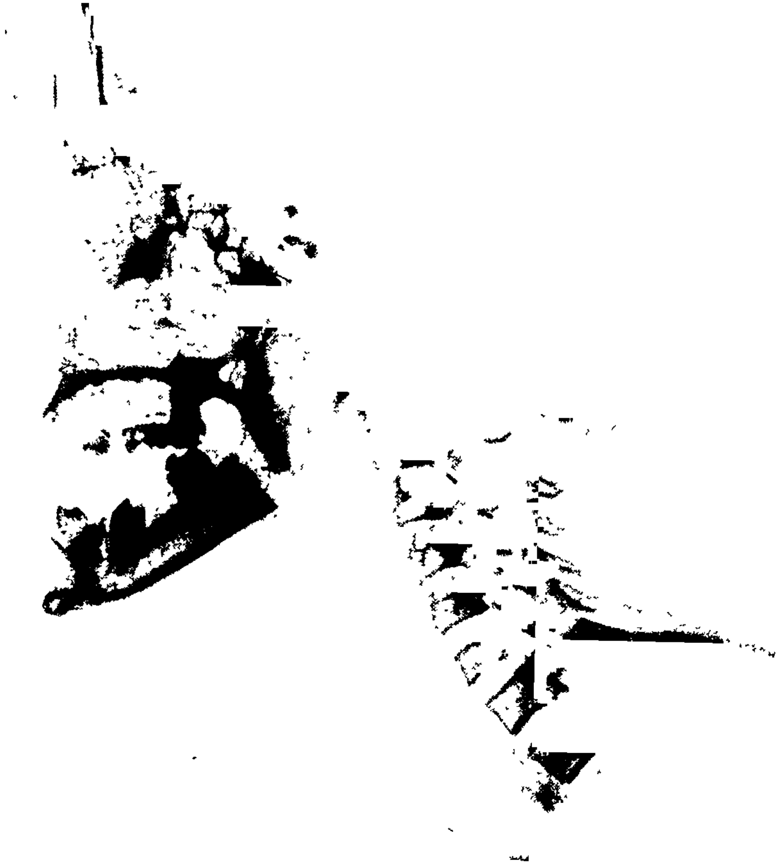


FIG. 6.

The same patient saying "s . . . s . . . s . . ." Practically no movement can be seen on saying "s . . . s . . . s . . ." (or on blowing). Incompetent sphincter action—gross nasal escape.

hypernasality. Tonsil beds and pharynx looked normal. The palate was very short and movement was absent. Both parents and the remaining four members of the family had normal throats. One sister had had tonsils and adenoids removed without any subsequent trouble.

Comment. A severe case of hypernasality; speech therapy started soon after operation might have restored palate movement and improved speech.

2. (*Case IV*)

A.A., aet. 4, was examined at the school clinic and advised removal of tonsils and adenoids on account of deafness, pain in the ears and nasal discharge No palatal abnormality was recorded Tonsils (guillotine) and adenoids were removed on 25 5 54 Severe post-operative bleeding from the adenoid bed was stopped by the insertion of a post-nasal pack Hypernasality developed immediately after operation Examination showed a very short palate with some tightening and inability to effect nasopharyngeal closure There was no scarring or distortion Speech therapy was commenced immediately Despite the return of good palate movement, hypernasality persisted and is still present The parents and one sister had normal palates, but the other sister showed definite shortening

Comment This boy s post-operative hypernasality was associated with a short palate, but was doubtless aggravated initially by poor movement of the soft palate following the insertion of a post-nasal pack It was hoped that an operation might help the nasal escape but the father was not keen and was inclined to be aggressive and litigious

3 (*Case VII*)

E P , aet 5, was referred by her general practitioner to the hospital clinic on account of sore throats mouth breathing and nasal discharge Her mother described her as a ' lovely speaker" before operation The removal of tonsils (dissection) and adenoids was followed by a severe nervous upset, when she ' never slept for months' Speech therapy for hypernasality was started one month after operation The soft palate appeared adequate and normal but there was slightly diminished movement on the left side Marked nasal escape was present on all sounds The child was nervous and the parental care was over solicitous The mother s throat was normal

Comment I could find no obvious organic cause for the hypernasality The very slight limitation of movement on one side of the soft palate seemed insufficient to account for the gross nasal escape The nervous child and severe psychological upset after operation led me to believe that this was a hysterical hypernasality of the type referred to by Froschels and Poe

4 (*Cases XII and XIII*)

M K and H K (twins) were seen at the school clinic on account of large tonsils , mouth breathing and "thick speech ' They were advised removal of tonsils and adenoids The operation (guillotine) was carried out at the age of 5 years Both children were hypernasal afterwards and have had intermittent speech therapy over the past 8 years Both still have slight nasal escape Examination showed no scarring or distortion and good palate movement The palate showed moderate shortening in one child and slightly less shortening in the other

Comment. Both twins had a pre-operative hyponasality converted to open nasal speech after operation. Response to speech therapy has been good.

Review of the Literature

The condition of "insufficiency of the palate" was first described in detail in a monograph by Lermoyez in 1892, although Roux had reported a case as early as 1825. Lermoyez described a congenital condition, in which the soft palate, although moving freely, does not reach the posterior pharyngeal wall and hypernasality results.

In 1910, Brown Kelly, in an excellent paper, gave the first detailed account of the condition in this country. He stated that the shortness of the palate was congenital, and measurements showed that shortness was present both in the hard and soft palates. He described two groups of cases associated with insufficiency, (*a*) those with submucous cleavage of the palate, and (*b*) those with muscular weakness due to defective development or abnormal disposition of the muscles of the soft palate. The cases he described developed hypernasality as soon as the child began to speak and the defect became less marked as the child grew older. He did not mention cases of latent hypernasality where the speech is normal until the tonsils or adenoids are removed. Indeed, he found the adenoids to be small in these cases. He also noted that the condition occurred frequently in patients below the average in intelligence and physical development. He regarded the insufficiency of the palate as one manifestation of general retarded development.

Since Brown Kelly's paper, numerous articles have been published on congenital palatal insufficiency without reference to the rôle played by the tonsils and adenoids in assisting nasopharyngeal closure.

In 1928, Poe described five children of the same family tree who developed hypernasality after tonsillectomy. The condition was transient in three children, but one had remained hypernasal for several years and the other was still hypernasal when he wrote the article six weeks after the operation.

Five years later, Poe published a further article describing additional cases and giving his opinion on the ætiology. His views were similar to those published by Fröschels (1925) at an earlier date. Both stressed the importance of post-operative pain in the causation of hypernasality. They stated that children feeling pain after operation, tend to expend the least possible muscular effort in speaking to save themselves unnecessary suffering. This leads to bad speaking habits with hypernasality, which, if not rapidly corrected become deep-rooted. Poe felt the condition was due to a "loss of proper innervation and co-ordination of the soft palate while speaking". While he suggested that removal of adenoids increased the difficulty of closing the nasopharynx, he made no mention of congenital insufficiency of the palate Although advising treatment by electrical stimulation, stretching, and massage of the palate, he considered speaking exercises most effective and advised frequent repetition of the words "appa" and "abba", spoken with gusto

Imre (1950) advised paraffin injection into the Passavant cushion in order to help closure of the nasopharynx Brown Kelly (1910) had previously considered this treatment, but did not think it likely to be of lasting value

Stern of Vienna suggested passing a tube from the nose to the ear so that the patient could hear the nasal escape for himself

In a recent article, Calnan (1957) pointed out the relationship of hypernasality to the removal of adenoids stressing the need for caution in advising adenoidectomy He did not consider tonsillectomy by itself likely to affect speech He suggested that some cases have a congenital deep pharynx rather than a congenital short palate

While textbooks on Speech Therapy recognize the importance of tonsil and adenoid removal in the causation of hypernasality, most ear, nose and throat textbooks fail to give the condition the attention it merits

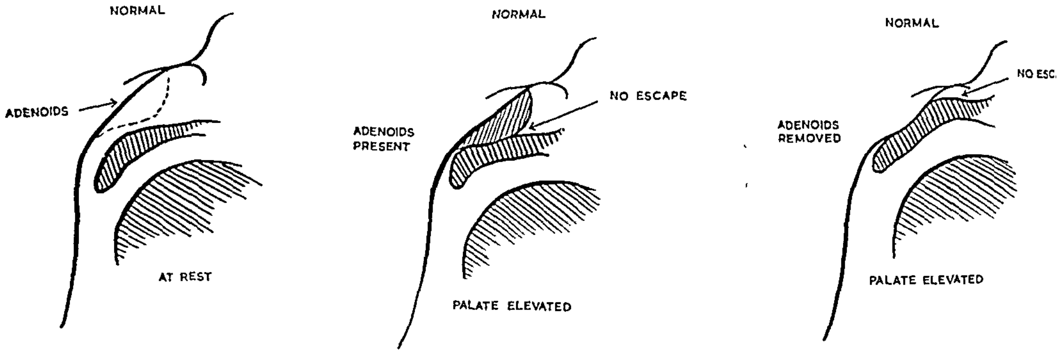
Ætiology

The underlying cause is shortening of the palate leading to an increased distance between the palate and posterior pharyngeal wall with incompetence of the palato pharyngeal sphincter The association with a cleft uvula and congenital defects elsewhere leads me to believe that the origin of the shortening is in all cases congenital Pseudo-shortening of acquired origin may be caused by a very large mass of adenoids pressing on the soft palate This has been referred to by Musgrove (1952) I removed the tonsils and adenoids in one such case with some trepidation thinking the palate to be congenitally short Instead of nasal escape developing I was surprised to find, contrary to Musgrove s observations, that the palate had returned to a normal position after operation and speech was unaffected The degree of congenital insufficiency varies considerably In some cases the palate shortening is so severe as to cause hypernasality even before the tonsils and adenoids are removed (Cases VIII, X, XI XVI) If operation is then undertaken, the hypernasality is greatly increased More frequently latent hypernasality is present, closure of the nasopharynx being possible only so long as the tonsils and adenoids are present If removal is carried out effective sphincter action is lost and nasal escape results

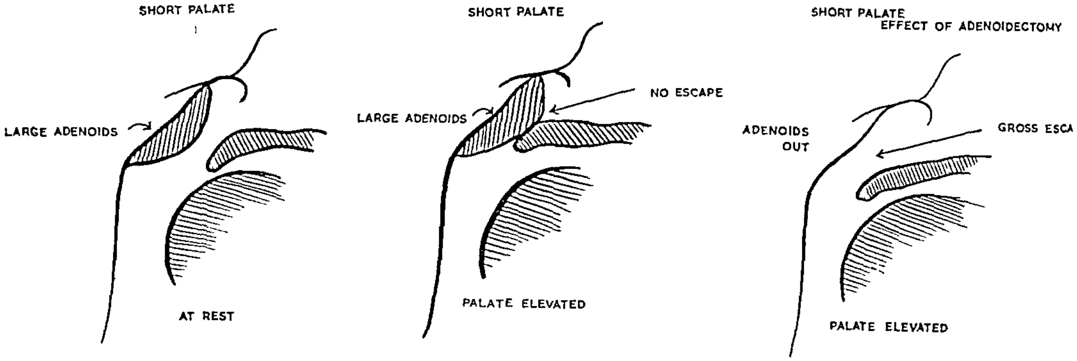
The mechanism of production is complicated and more than one factor is normally responsible

1 *Effect of removal of adenoids* A large mass of adenoids by virtue of its bulk helps in closure of the palato pharyngeal sphincter When the

adenoids are removed, the nasopharynx is deepened and the sphincter mechanism may no longer remain effective (Figs. 7-12).



FIGS. 7, 8, 9.
Normal palate showing effective closure of nasal airway.



FIGS. 10, 11, 12.
Case of palatal insufficiency: effective sphincter action is lost after adenoidectomy.

2. *Effect of removal of tonsils.* (*a*) Removal of tonsils leaves a raw area in the soft tissues of the tonsil fossae which heals by fibrosis. The fibrous tissue contracts giving a slight tightening effect on the palate which tends to draw it forward and deepen the nasopharynx. (*b*) Very large tonsils may push the posterior pillars of the fauces backwards carrying the palate with them thus helping to close the nasopharynx: when these tonsils are removed the posterior pillars and palate move forward, and increase the depth of the nasopharynx (Fig. 13).

3. The operation may result in temporary immobility of the soft palate. Sometimes, as Poe (1933) suggested, this is due to post-operative pain, while in one case in this series, the insertion of a post-nasal pack seemed an important factor. If palate movement is not rapidly restored, nasal escape may result.

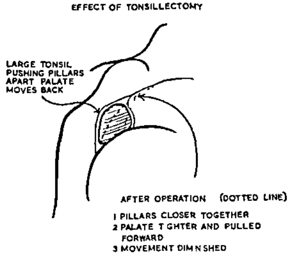

EFFECT OF TONSILLECTOMY
LARGE TONSIL PUSHING PILLARS APART PALATE MOVES BACK
AFTER OPERATION (DOTTED LINE)
1 PILLARS CLOSER TOGETHER
2 PALATE TIGHTER AND PULLED FORWARD
3 MOVEMENT DIMINISHED

FIG 13

Effect of tonsillectomy, illustrating the slight forward displacement of the soft palate following operation

4 The nervous upset of the operation may result in hysterical hypernasality This may start with immobility of the palate due to pain, but nasal escape persists after palate movement is restored Gutzmann referred to this as a "habit palsy" (Gewohnheitslahmungen) It is easy to see how a hysterical case may arise, for most normal individuals can readily imitate cleft-palate speech Some cases of organic origin may well have a functional overlay This may be aggravated if legal action is contemplated

The association with retarded mental development as mentioned by Brown Kelly was borne out in this series of cases

Conclusion The underlying cause in hypernasality is a congenital palatal insufficiency This defect, however, may sometimes be overcome by excellent palate movement (This would explain cases occasionally encountered in which there is marked palate shortening but no nasal escape) Any limitation of palate movement, such as frequently follows tonsil and adenoid operations, leads to weakened sphincter action In a palate of normal length the sphincter mechanism is unlikely to be seriously affected by this, but if there is the slightest degree of palate shortening, nasal escape is certain to result Once nasal escape is firmly established, it is extremely difficult to eliminate

Prevention

The question which faces every Ear, Nose and Throat Surgeon is not "How do I treat this condition?", but rather, "How can I prevent this condition occurring?"

Most surgeons are well aware of the dangers of tonsil and adenoid removal in cases of cleft palate They do not seem to be equally conscious of the dangers in patients with a short palate

The following unfortunate cases are good examples of what may happen:

1. P.B., aet. 4, was referred to the hospital clinic on account of "bad speech". She was advised removal of tonsils and adenoids. Operation was followed by a marked aggravation of the pre-existing defect. This child has now gross hypernasality.

2. J.M., aet. 5, attended the speech therapy clinic on account of dyslalia. The therapist noticed slight hypernasality in addition to the dyslalia and warned the mother of the possible effects of removal of tonsils and adenoids; the advice went unheeded or misinterpreted for the child returned minus tonsils and adenoids, but suffering from gross hypernasality. "If she blows a balloon, she does much better if she holds her nose", was the apt description of the mother.

By contrast, the harmful effects of operation were avoided in the following case:

3. A.T., aet. 11, female, was referred for tonsillectomy on account of "repeated sore throats". Her tonsils were large, and she had a tonsillar gland on the left side. It was noticed that her palate was very short (+++) and she had a double uvula. Marked mental retardation was also present. Nasal escape was present, especially on vowel sounds. She was accordingly referred to a speech therapist who found "a considerable degree of hypernasality" in the speech. On closer questioning, it was found that this girl had suffered from tonsillitis twice only. She was kept under observation for six months and, having suffered from no further sore throats during that time, was then discharged.

Comment. I am convinced that this girl's hypernasality would have been considerably aggravated by removal of tonsils. This would indeed have been a high price to pay for relief from occasional sore throats.

The surgeon must, therefore, firstly avoid operations on cases already hypernasal. The opinion of a speech therapist should be sought in any doubtful case. Secondly he must watch carefully for cases of latent hypernasality, where speech is normal, but the palate is short and the distance between the soft palate and the posterior pharyngeal wall is increased. His attention may be drawn to this possibility by a bifid or double uvula, but not all cases of cleft uvula are associated with a short palate. The assessment of the palate may be rendered difficult by (*a*) a refractory child, or (*b*) very large tonsils which meet each other and prevent an adequate view of the pharynx. If there is hypernasality already, or if the palate is short, operation must be avoided if at all possible. If operation is still thought to be essential, consideration should be given to:

(*a*) Removal of either tonsils *or* adenoids—not both.

(*b*) Removal of tonsils and later adenoids as a two-stage procedure.

(*c*) In cases of middle ear disease such as catarrhal deafness, the application of radium to the nasopharynx may avoid the need for adenoidectomy If adenoid removal is essential the lateral adenoid masses may be removed with preservation of the central pad (Calnan)

(*d*) Speech therapy and exercises before and immediately after operation

(*e*) Adequate sedation for a nervous child

(*f*) Finally, the parents must be warned of the possible consequences It is wise to get a written statement signed by one of the parents as a safeguard against legal action

Treatment

The general principles of treatment in this condition are similar to those applied to cleft palate cases For this reason I do not propose to deal with the treatment in detail for it must involve close co operation between surgeons and speech therapists The primary responsibility for starting and directing treatment must nevertheless remain with the throat surgeon who performed the operation It is important for him to appreciate both the seriousness and the urgency of the condition and start treatment before the child leaves hospital

There are three main aims in treatment

(*a*) Strengthening of palate muscles and restoration of full movement

(*b*) Elimination of nasal escape

(*c*) Correction of faults in articulation (if these exist)

The patients fall into two groups

I Cases with lesser degrees of palate shortening where re establishment of good movement eliminates nasal escape

II Cases of severe shortening which are likely to have persistent nasal escape even with good palate movement

It is usually easy from the clinical examination to decide into which group a case will fall but in cases of doubt radiographic study may be found helpful in showing the degree of incompetence present

Treatment in Group I Cases

1 Active exercises to strengthen palate muscles and restore movement (It should be kept in mind that there may be a congenital weakness of the muscles of the soft palate)

(*a*) *Blowing exercises* are commonly used, for, in addition to producing an active palate contraction, they also give practice in directing the breath through the mouth and away from the nose. The exercises can be made interesting by blowing up balloons, blowing out candles or games of blow-football.

(*b*) *Suction exercises* produce strong active contraction of the soft palate muscles. All drinks should be taken through a straw, while more difficult exercises can be given, such as sucking up peas or other objects by means of a straw.

The alternate blowing and sucking used in playing a mouth organ provide an excellent, if somewhat noisy, method of strengthening the palate muscles.

(*c*) *Frequent repetition of plosives.* The following sounds, pronounced energetically, are useful in providing rapid palate contraction, pm, tn, kng, pk, abba, appa.

These exercises are best carried out by a speech therapist who must be firm and persuasive, for slovenliness and lack of effort lead to a persistence of nasal escape.

2. Correction of any errors in articulation.

Treatment in Group II Cases

These cases require operative treatment in order to eliminate nasal escape, due to the gross insufficiency of the palate.

1. *Palate operations.* If the palate is very short, one of the many forms of palate lengthening operations is indicated.

2. *Pharyngoplasty.* In less severe cases a pharyngoplasty operation of the Wardill type to increase Passavant's cushion, has the effect of replacing the adenoid mass which has been removed. If more efficient closure is required, a Rosenthal pharyngoplasty, which achieves marked narrowing of the isthmus, may be effective. The most severe cases are likely to require both palate lengthening and pharyngoplasty.

Intensive speech therapy and exercises should follow all operations.

Additional forms of treatment

1. Electrical stimulation may be helpful to keep up the tone of the muscles of the soft palate.

2. Relaxation exercises and psychiatric treatment may be required in nervous or hysterical cases.

3. Digital massage of the palate is unsuitable in this type of case. It would merely tend to focus the child's attention on the disability. It should be reserved for cases of scarring.

Summary

Hypernasality (Rhinolalia aperta) is an uncommon complication of operations for removal of tonsils and adenoids An investigation carried out in the Eastern and part of the South Eastern Regions of Scotland revealed 19 cases operated on by 11 different surgeons in a total of 27 734 operations In none of these cases was post operative scarring present The ætiology is discussed and congenital insufficiency of the palate together with diminished post operative palate movement is considered to be the usual cause Early recognition and immediate treatment are essential

Acknowledgments

I wish to thank all those who assisted me in this Survey especially those surgeons who allowed me access to their cases and the Speech Therapists who gave of their time unsparingly I am also indebted to Professor A Mair who helped in the initial planning of the Survey to Mr Wm Malin for his diagrams and to Mr H D Brown Kelly for his translations

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PAPILLOMA OF THE LARYNX: A CLINICO-PATHOLOGICAL STUDY

By P. WINSTON and S. S. EPSTEIN* (London)

Papilloma of the larynx although rare, is by far the commonest benign laryngeal neoplasm. Capps (1957) on the basis of a survey of leading centres in England recently stated, that "the majority of clinics are likely to deal with less than 10 cases in a 5-year period". Nevertheless as pointed out recently by Epstein, Winston, Friedmann and Ormerod (1957), they represent some 10 per cent. of all benign vocal cord "tumours"—this term being used to include all neoplastic and non-neoplastic swellings.

It is our object to record here the incidence of this condition and to assess its clinico-pathological implications with particular reference to the problem of malignant change. This paper forms the third and final part of a systematic study of benign laryngeal "tumours", based on recent material seen at the Royal National Throat, Nose and Ear Hospital, London (Epstein and Winston, 1957; Epstein *et al.*, 1957).

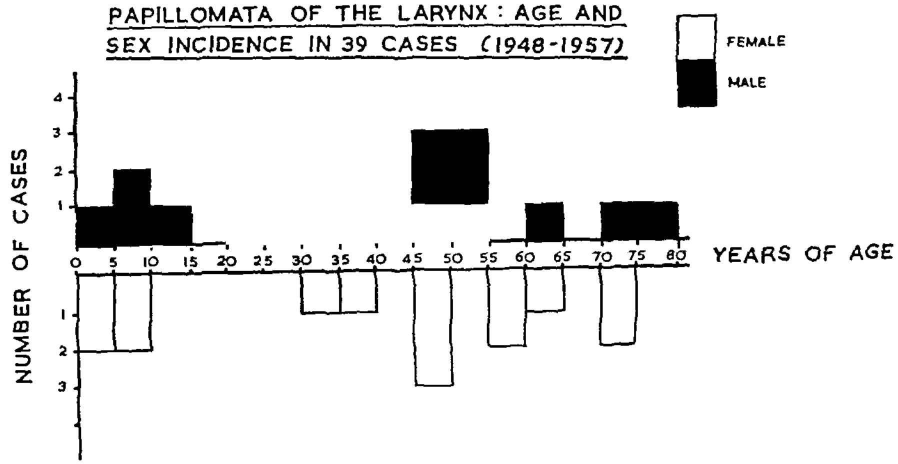

Fig. 1

Incidence

Fig. 1 presents an analysis of the age and sex incidence of 39 papillomata studied over a 10-year period (1948-1957). As can be seen, 31 cases presented in adult life and 8 under 15 years of age There is an equal sex distribution in the latter, whereas males are twice as common in the former This greater propensity of the adult male has been noted previously by many authors including Crowe and Breitstein (1922) and Bjork and Weber (1956)

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Ætiology

The striking differences in the behaviour and natural history of adult and childhood varieties of laryngeal papillomata, may indicate a different ætiology The latter by reason of its multiplicity, tendency to spontaneous regression and response to the tetracyclines alleged by a variety of workers, including Holinger *et al* (1950) and Zakrzewski and Soboczynski (1957), has by analogy with the common infective wart suggested a virus ætiology Childhood papillomata, however, are not familial and do not appear to be contagious Bjork and Weber (1956) nevertheless claim to have found a significant correlation between the incidence of these and common warts in any one individual Yet apart from some experimental work by Ullmann (1923), who demonstrated transmission of childhood papillomata by inoculation of "cell free filtrates", no further objective evidence of a virus ætiology has been advanced More recently preliminary tissue culture studies by Brewer and Kalter (1957), have shown the presence of inclusion bodies in the juvenile papilloma, but their significance remains to be assessed The alleged regression at puberty of this type and the response to local œstrogens claimed on the basis of a small series of cases by Broyles (1941), may suggest the influence of hormonal factors without necessarily incriminating them ætiologically Bjork and Weber (1956) critically examined the question of puberty regression and point out that the statistical evidence for this is tenuous Certainly in their series of 37 children, puberty seems to have had little effect on the natural history of this disease In the 8 children reported in this series, only 3 have been followed until after puberty and in all cases the neoplasms have persisted In the remaining 5 puberty has not yet supervened It is of interest in this connection to recall the description by Holinger *et al* (1950) of an adult female, whose papillomata temporarily regressed with each of her successive 3 pregnancies

The possible ætiological role of dietary factors has of late attracted attention Bjork and Weber (1956) noted a striking increase in the incidence of papillomata in Finland during the war and early post war years, when the local population was subjected to severe dietary deprivations Similar findings were also noted in Holland during the recent occupation Furthermore Messerklinger (1956) reported a dramatic response of childhood papillomata to the oral administration of methionine—an essential amino acid and lipotrophe He suggested that a disturbance of methionine balance, or of fat metabolism, may be an ætiological factor

Tapia (1956) noted a high incidence of papillomata in Mexico, where they comprise 67 per cent. of all benign laryngeal "tumours". It is difficult to evaluate the precise rôle of dietary factors in this demographic pattern.

Histology

A papilloma is a benign epithelial neoplasm, consisting of irregular villous processes composed of thin branching fibro-vascular fronds, covered by a well differentiated epithelium. A distinct basement membrane separates the epithelium from its subjacent stroma, in which an occasional and irregularly distributed infiltrate of lymphocytes may be seen. The basal cells of the epithelium are orientated radially with reference to the long axis of their parent frond and the more superficial cell layers—tangentially. The laryngeal papilloma is essentially a squamous neoplasm in which keratinization may occur. Hyperkeratosis (Fig. 2) in fact was found to be unusual in our material; parakeratosis on the other hand was commonly seen (Fig. 3). The neoplastic process may sometimes involve respiratory epithelium and occasionally the juvenile papilloma presents composite areas, where both squamous and respiratory epithelia are adjacently represented (Fig. 4).

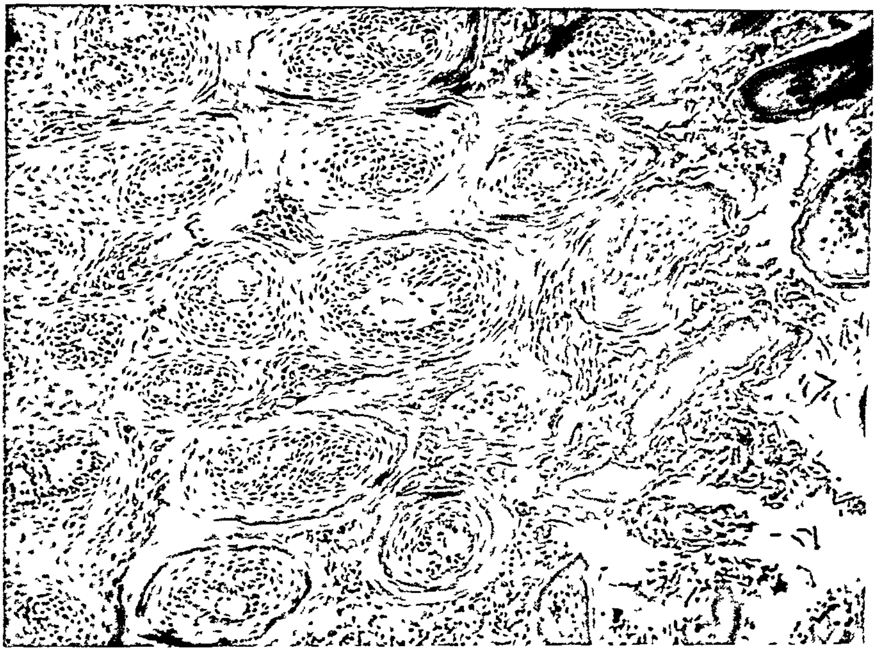


FIG 2

Hyperkeratotic papilloma (× 125)

Histologically it is usually easy to recognize a papilloma as conforming to the above description, although as will be pointed out subsequently it

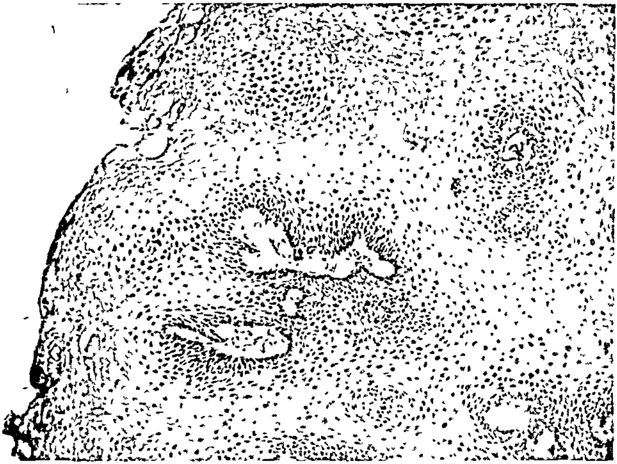


FIG 3
Papilloma exhibiting parakeratosis (× 155)

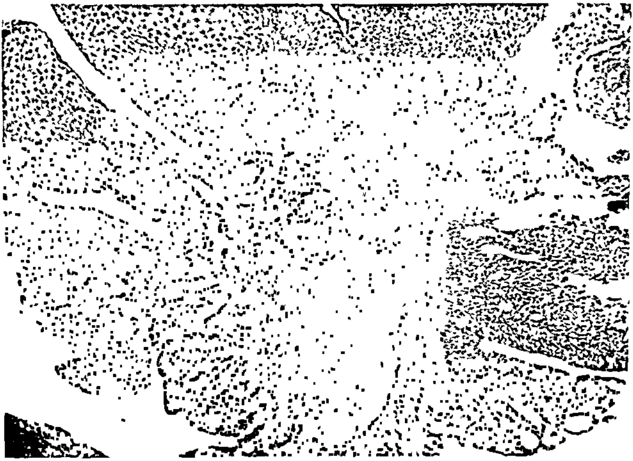
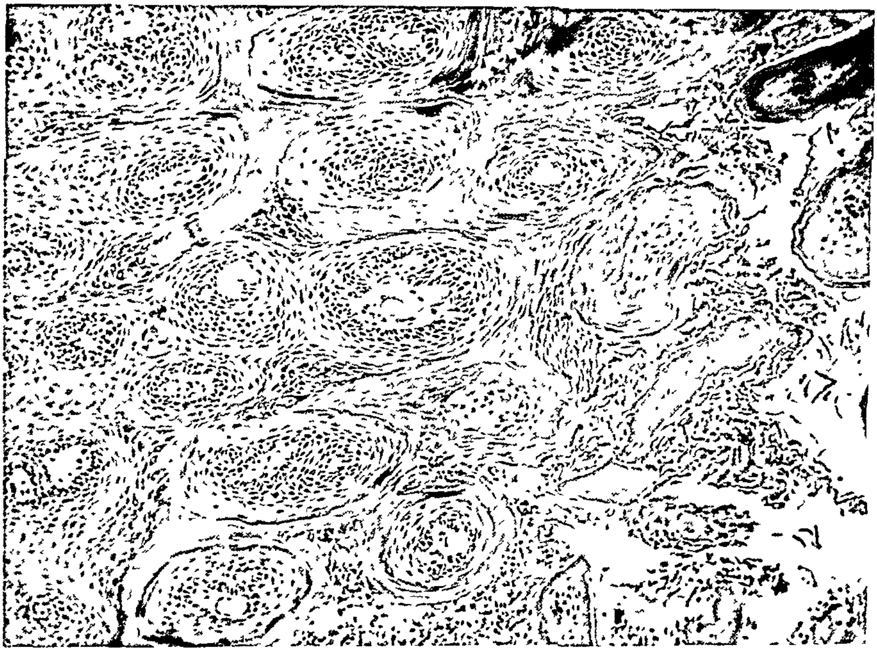


FIG 4
Juvenile papilloma involving both squamous and respiratory epithelia (× 135)

Tapia (1956) noted a high incidence of papillomata in Mexico, where they comprise 67 per cent. of all benign laryngeal "tumours". It is difficult to evaluate the precise rôle of dietary factors in this demographic pattern.

Histology

A papilloma is a benign epithelial neoplasm, consisting of irregular villous processes composed of thin branching fibro-vascular fronds, covered by a well differentiated epithelium. A distinct basement membrane separates the epithelium from its subjacent stroma, in which an occasional and irregularly distributed infiltrate of lymphocytes may be seen. The basal cells of the epithelium are orientated radially with reference to the long axis of their parent frond and the more superficial cell layers—tangentially. The laryngeal papilloma is essentially a squamous neoplasm in which keratinization may occur. Hyperkeratosis (Fig. 2) in fact was found to be unusual in our material; parakeratosis on the other hand was commonly seen (Fig. 3). The neoplastic process may sometimes involve respiratory epithelium and occasionally the juvenile papilloma presents composite areas, where both squamous and respiratory epithelia are adjacently represented (Fig. 4).



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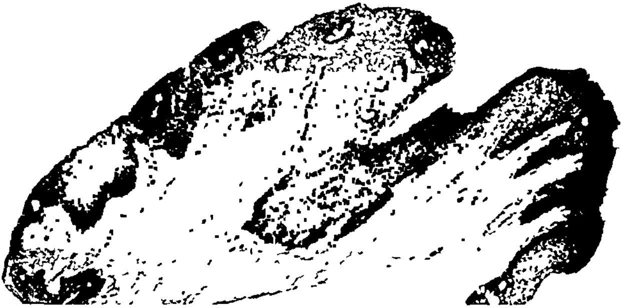

Fig 7
Papillary keratosis epithelial infolding has produced a picture with superficial similarity to a papilloma (× 35)

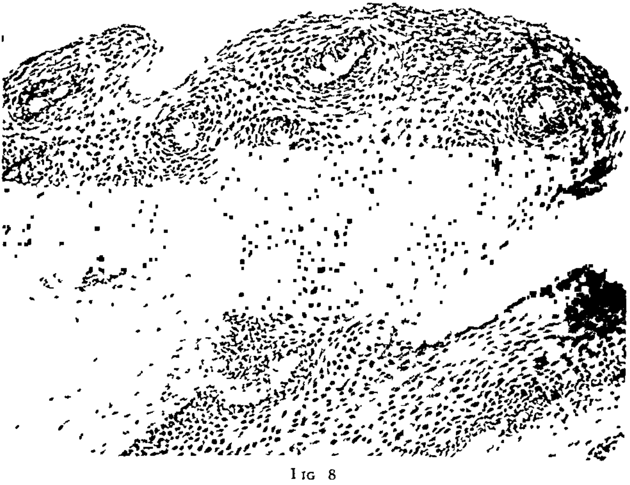

Fig 8
Papillary keratosis further detail of Fig 7 (× 200)

may present in different clinical guises. Conversely the clinical diagnosis of papilloma may not always be substantiated in the laboratory. Occasionally and especially if the biopsy is small, it may be difficult in the adult to differentiate the papilloma from a papillary keratosis—the latter term being applied to a heaped-up papillary lesion arising from a keratotic epithelium. This heaping up can produce frond-like structures which can (Figs. 5, 6, 7 and 8) superficially resemble the papilloma. Critical examination, however, usually reveals that the typical organization and relation of epithelium to stroma is lacking. Moreover there is usually a well marked stromal infiltrate subjacent to the keratotic epithelium.

Malignant Change

The outstanding histological problem of the laryngeal papilloma, is the definition and recognition of early malignant change, especially in the recurrent adult type. It is possible to elaborate definitive criteria for the recognition of *pre-invasive cancer* or *carcinoma in situ*, such as basal cell hyperactivity and atypism extending through the whole thickness of the epithelium. Nevertheless in the ultimate analysis there is, as indicated by occasional differences between pathologists over the interpretation of a given section, a significant subjective element in this diagnosis. It is known that an epithelium can react to trauma—say of a biopsy—by subsequent rapid and ill-organized regenerative growth. Local infection can also induce similar reactive changes. The implication of these changes, must further be qualified by reference to such considerations as the site of the epithelium and the age of the patient. Altmann *et al.* (1955) have suggested that the significance of *carcinoma in situ* is less serious in a papilloma, than in a flat laryngeal epithelium.

Furthermore in the present series, foci of epithelial changes have been detected in papillomata of 2 childhood cases on occasions during their serial protracted removal, which had they occurred in adults may well have been regarded as *carcinoma in situ*. In none, however, did these changes persist and subsequent biopsies revealed no persistent atypism.

TABLE I

THE INCIDENCE OF MALIGNANT CHANGE IN LARYNGEAL PAPILLOMATA TO DEMONSTRATE WIDE DIVERGENCE OF OPINION AMONG VARIOUS AUTHORS

Author	Year	No of Papillomata in series	Per cent incidence malignant change
Jackson and Jackson	1937	205	3
New and Erich ..	1938	194	1·5
Cunning	1950	276	14
Holinger *et al.* ..	1950	109	0
Altmann *et al.* ..	1955	51	21·5
Putney	1955	501	2
Amarante	1957	36	20

The incidence of malignant change in the adult papilloma has been variously assessed. Table I based on data presented by 7 authors is representative of the wide divergence of opinion that exists on this subject. Further examination of this data is of interest In the series reported by Jackson and Jackson (1937), all cases occurred in adults, whereas half of those analysed by Holinger *et al.* (1950) were children. The heterogeneity of such data is, as emphasized by Altmann *et al.* (1955), further increased by the inclusion of established papillary carcinomata Moreover the criteria of malignancy in different series are by no means uniform. Malignancy in the 11 cases reported by Altmann *et al.* (1955) was determined on the basis of the recognition of "intra-epithelial anaplastic change" and in none was there evidence at any stage of invasive carcinoma In other series however, malignancy was not diagnosed in the absence of invasion In view of these difficulties, it is probably unwise at present to attempt to quantitate on the frequency of this hazard.

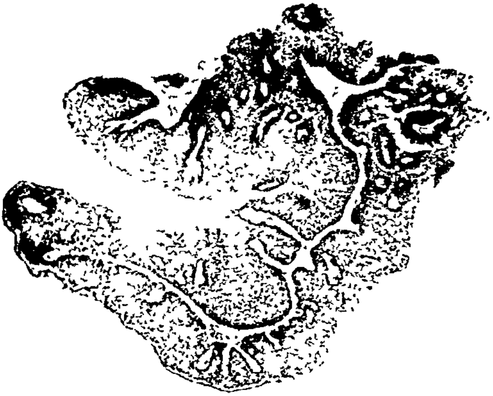

FIG 9
Benign adult papilloma (× 65)

In the present series of 31 adult cases, it has been necessary to consider problems of malignancy in 4 instances In only one of these, however, was there unequivocal evidence of *carcinoma in situ* developing in a previously benign squamous papilloma (Figs 9, 10 and 11) In a further case changes developed which while not constituting *carcinoma in situ*, were so suspicious that it was decided to regard these as such Both of these cases

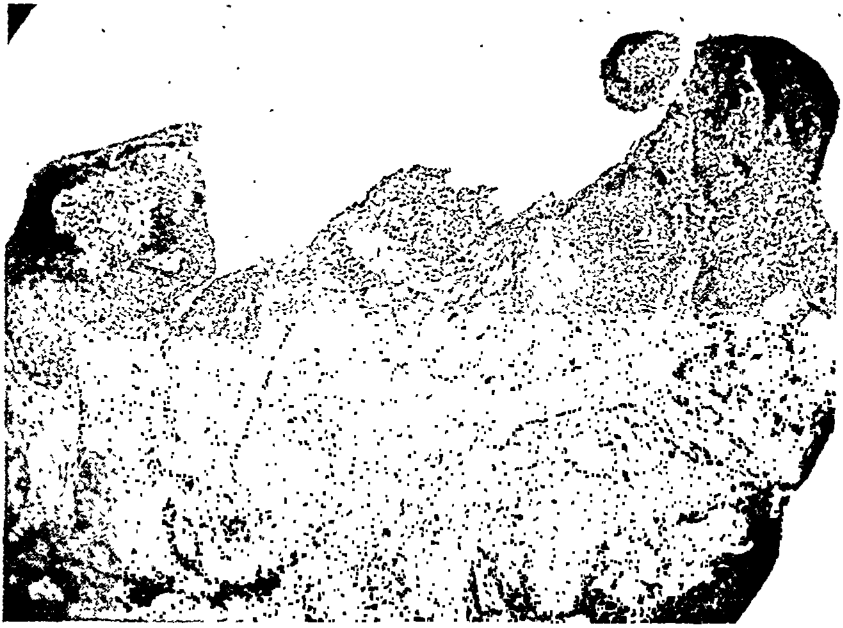

FIG. 10

Malignant change in papilloma of previous patient 1 year later (× 65) See Fig. 9.

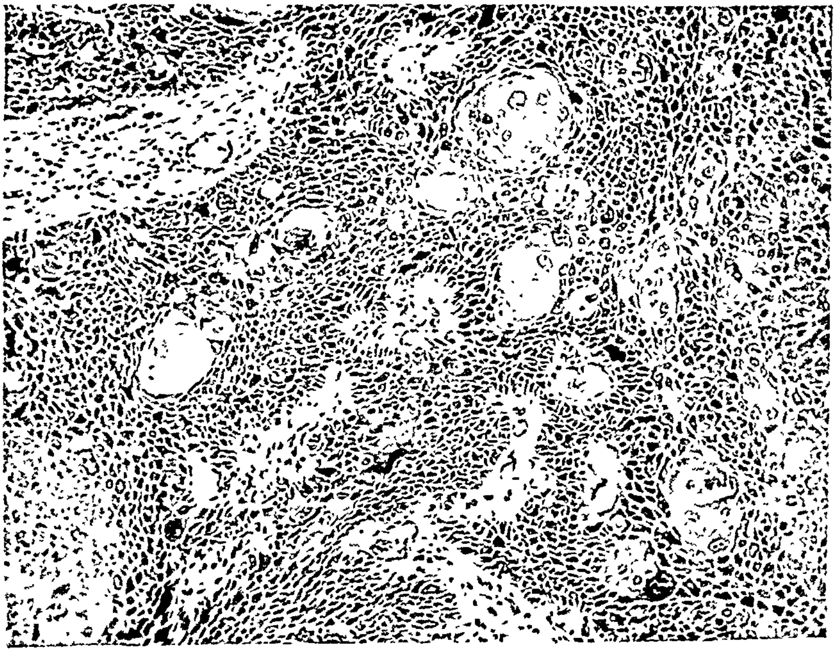

FIG 11

Malignant change in papilloma further detail of Fig 10 (× 265) were successfully treated by local removal and irradiation In a further 2 instances initial biopsies revealed areas of *pre-invasive cancer* It must however, be appreciated that these may have represented well differentiated papillary carcinomata *de novo* rather than papillomata, in which malignancy supervened Laryngofissure was performed with success in both

Symptoms

In the majority of both adult and childhood cases, symptoms were those of voice change and ranged from slight dysphonia to complete aphonia In 6 of the latter, elective tracheotomy was performed for respiratory obstruction

Signs

Whilst individual neoplasms in all adults of this series were localized to the true cords those in childhood had a wider distribution—extending in some instances from the epiglottis to the trachea All 8 childhood cases were multiple and recurrent, the numbers—both of neoplasms and recurrences—often being extremely high The adult papillomata are analysed with reference to recurrence and multiplicity in Table II

TABLE II

ADULT PAPILLOMATA OF THE LARYNX AN ANALYSIS OF 31 CASES IN RELATION TO MULTIPLICITY AND RECURRENCES

Single non recurrent	15
Multiple non recurrent	6
Multiple recurrent	5
Single recurrent	5

As can be seen approximately 33 per cent were multiple but in contrast to those of childhood, the number of individual "tumours' rarely exceeded 3 Holinger *et al* (1951) found a similar incidence of multiplicity in adults but Altmann *et al* (1955) reported an incidence of only 25 per cent It can also be seen from Table II that 33 per cent of the adult cases were recurrent and among these are numbered both single and multiple varieties Recurrence of either type may be due to the development of further "tumours" from adjacent epithelium, or regrowth at the original site probably due to incomplete removal The risk of malignant change tends to be higher in recurrences

Childhood and adult papillomata commonly present identical appearances They are usually pedunculated—though occasionally sessile—fronded, vascular and friable This appearance is, however by no means pathognomonic and as indicated in Table III, they may be simulated by non-neoplastic lesions such as polyps Conversely the conditions enumerated in Table IV, may themselves be simulated by the papilloma

TABLE III.

ORIGINAL CLINICAL DIAGNOSIS OF 39 CASES OF HISTOLOGICALLY PROVEN PAPILLOMATA.

A. Papilloma		25
B. Other clinical diagnoses		14
Polyps	5	
Chronic laryngitis	3	
Contact ulcers	2	
Carcinoma	3	
Granuloma	1	

TABLE IV.

CASES CLINICALLY DIAGNOSED AS PAPILLOMATA, BUT SUBSEQUENTLY HISTOLOGICALLY DIAGNOSED AS FOLLOWS:

Polyps	21
Papillary keratoses	4
Non-specific granuloma	2

Treatment

The commonest mode of treatment in our series was simple removal. In childhood this often required repeated removal on frequent occasions. The necessity for this does not seem to have been diminished by the use of tetracyclines, testosterone or local application of silver nitrate or podophyllin—the latter as advocated by Hollingsworth *et al.* (1950) and more recently by De Vido and Megighian (1957). It must be clearly appreciated that while the juvenile papilloma is histologically a benign condition, yet clinically its implication may be no less serious than that of cancer. Crowe and Breitstein (1922) pointed out that the mortality in children under the age of 5, is probably greater than that of carcinoma of the larynx in adults. Ferguson and Scott (1944) again emphasized the fallacy of regarding this condition as clinically benign. The possibility of fatal asphyxia, hæmorrhage or aspiration bronchopneumonia is very real. Of the 8 cases reported here, there were 2 fatalities—one from asphyxia and the other from secondary hæmorrhage. Tracheotomy was performed in 6 childhood cases for the relief of dyspnœa and never merely to *rest the larynx* as practised by Mackenzie (1901) and de Sanson (1957). Routine adoption of prophylactic tracheotomy, however, would probably eliminate a major hazard to life in papillomatosis of childhood. Pulmonary implantation metastasis reported by Hitz and Oesterlin (1932) and Kirchner (1951), is a rare complication that was not noted in our series. The possibility of malignant change in the papilloma of childhood cannot be entirely discounted. Walsh and Beamer (1950) reported this occurrence in 2 children, aged 12 and 13 years respectively. The application of low doses of radiation, as recommended by New (1921) and Foster (1933), has not yet been performed in any of our cases, although one child did in fact receive deep X-ray therapy elsewhere prior to being referred to this centre. The use of irradiation for non malignant conditions has been further discouraged by the recent findings of Simpson, Hempelmann and Fuller

(1955), who reported an enhanced incidence of thyroid carcinoma in patients whose necks had been irradiated in childhood

The problem of the adult papilloma is very different In addition to the maintenance of phonation and an adequate airway, the possibility of malignant change—though small—is real and demands a more radical approach The histology of the recurrent adult neoplasm must be carefully assessed Altmann *et al* (1955) recommend a conservative approach in the absence of histological evidence of invasion On the basis of careful observation of 51 adult cases they concluded that the significance of epithelial atypism is less grave here, than similar changes occurring in a flat keratotic laryngeal epithelium It is the consensus of modern opinion, however, that should there be any suspicion of early malignant change even of a pre-invasive character, laryngofissure and excision of the affected cord are indicated In our series this was performed twice 2 further patients received radiotherapy for the same reasons The former cases have been followed up for 3 and 4 years the latter for 4 and 9 years respectively and there has been no recurrence of carcinoma It is, of course, appreciated that this success may in some instances be apparent only and constitute a reflexion of an unduly rigorous interpretation of the initial histological appearances

Summary

The papilloma is by far the commonest benign laryngeal neoplasm A series of 39 cases of laryngeal papillomata, of which 8 occurred in childhood and 31 in adult life, have been reported Data on the ætiology of this condition have been reviewed and it is suggested that the importance of dietary factors has been underestimated Childhood papillomata are usually diffusely distributed, multiple and recurrent The adult neoplasm is commonly localized to the true cords and less often multiple and recurrent It is indicated that the evidence for puberty regression of the childhood type is tenuous The possibility of fatal asphyxia in children with laryngeal papillomata is real but may in some instances be reduced by the adoption of prophylactic tracheotomy The histology of the adult papilloma must be carefully assessed to detect early malignant change, unequivocal evidence of which occurred in only one instance in the series reported here

Acknowledgments

The authors would like to express appreciation to Professor F C Ormerod and Dr I Friedmann for their kind help and advice They would also like to thank Miss V Shepherd for sections Mr D Connolly for photomicrographs Mrs T Woof for assistance with the bibliography and finally the surgical staff of the Royal National Throat Nose and Ear Hospital for permitting access to their case records

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THE TRANSVERSE DIMENSIONS OF THE NASOPHARYNX IN CHILD AND ADULT WITH OBSERVATIONS ON ITS CONTRACTILE FUNCTION

By W STIRK ADAMS (Birmingham)

DURING recent investigations of the value of irradiation of the nasopharynx in tubotympanic dysfunction with the M R C 50 mg radium applicator, the calculation of the r dosage received at different positions in the tissues of the nasopharynx required detailed measurement of its spacial anatomy

From standard tables the r dosages at various distances from the 50 mg radium applicator used in these treatments, are readily ascertained, but a record of the dimensions of the space at different ages in early childhood was not available, though a previous study of these dimensions had been made by Remaggi in patients over the age of 8 years

In this, the observations were made during a research which was undertaken to ascertain whether a chronic pharyngitis of atrophic or hypertrophic type might be associated with a reduction or expansion of the normal dimensions He measured *in vivo* the antero-posterior and transverse diameters of the nasopharynx in 179 normal individuals from 8 years old to the seventh decade, and compared them with similar measurements in 51 cases of chronic pharyngitis

An analysis of his figures indicates that the antero posterior diameter of the nasopharynx increases as a hyperbolic function of age, and examination of his tables shows that little change occurs in this dimension with increasing years until late in life but the transverse diameter between the lateral recesses ranged from 3 19 cm in younger patients to 5 9 cm with a linear increase in the graph from an average of 4 35 cm at 8-9 years of age to 4 85 cm in the seventh decade

For the calculations of the r dose received at the pharyngeal orifice of the eustachian tube and outwards along the antero-lateral wall of the nasopharynx to the lymphoid tissue in the lateral recess, we required a detailed study of the lateral dimensions of the nasopharynx in young children, and in the present survey the measurements have been made in patients under a general anæsthetic for surgical procedures, and usually at the completion of a tonsil and adenoid operation In some of the earlier cases an unexpected degree of muscular contraction was encountered in the upper nasopharynx during phases of light anæsthesia, and the series of observations was extended for further investigation of this contractile function.

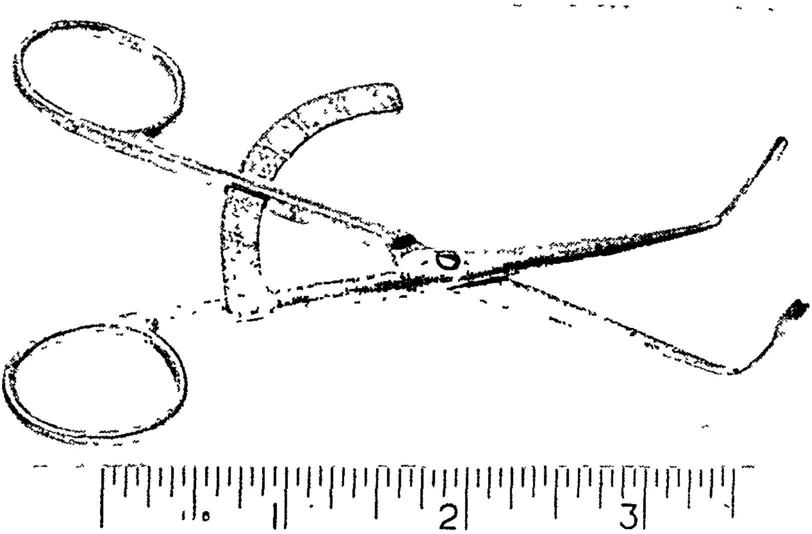

FIG. I.
The measuring gauge.

The Measuring Gauge (Fig. 1)

The gauge was designed and constructed by Messrs. Philip Harris & Company of Birmingham. It has a forceps structure with angled extremities, and a measuring arc has been attached to the shaft, calibrated in centimetres, which indicates the distance between the forceps points as they separate on opening.

Method of Use

In making measurements the forceps ends are introduced through the open mouth past the soft palate into the post nasal space. They are then opened widely to allow the arms to position themselves on the walls of the lateral recesses without undue pressure. The diameter is then read on the gauge. In the earlier cases digital control was required in positioning the arms, but with practice it was found that a gentle rocking movement allowed the opened forceps arms to position themselves in the lateral recesses. In measuring the distance separating the pharyngeal orifices of the eustachian tubes, however, digital guidance is necessary and the examining finger in the post nasal space identifies the tubal orifices, and the forceps arms are then placed in contact with these and the gauge reading is recorded.

TABLE I

TRANSVERSE DIAMETER OF NASOPHARYNX AND INTERTUBAL DISTANCES IN AGE GROUPS

Years 2–3			3–4			4–5			5–6			6–7		
Sex	Tv diameter	Inter tubal distance	Sex	Tv diameter	Inter tubal distance	Sex	Tv diameter	Inter tubal distance	Sex	Tv diameter	Inter tubal distance	Sex	Tv diameter	Inter tubal distance
									M	4 3	2 2			
M	4 3	1 9	M	4 4	—	M	5 2	—	M	4 2	—	M	5 0	—
	3 7	2 1		4 8	—		4 0	—		4 8	—		5 0	—
F	3 8	1 8		4 8	2 4		3 75	—		4 2	—		5 0	—
	4 2	1 9		4 8	2 2		3 5	2 5		5 4	—		4 8	—
	4 3	—		3 5	—		4 5	2 0		4 8	2 7		4 3	2 0
				4 4	2 0		4 6	2 2		5 0	2 6		4 3	—
				3 75	2 5		4 6	2 1		5 0	2 4		5 7	2 6
				4 8	2 4		4 0	—		4 7	2 6		5 0	2 4
				4 4	2 4		4 0	2 0		5 0	—		4 7	2 2
				4 2	2 0		4 7	2 1		4 0	2 5		4 9	2 4
				4 2	2 2		4 0	2 2		4 2	2 5		4 1	2 0
				4 1	2 1		4 9	2 2		5 2	2 8		4 5	2 5
				3 8	2 1		4 0	1 8		4 5	2 0		4 2	2 2
			F	4 2	2 2	F	4 8	2 8	F	4 8	—		5 2	2 4
				4 2	2 1		4 6	2 2		4 7	—		5 0	2 0
				3 2	2 4		4 5	—		4 5	—		4 7	2 3
				3 7	2 2		4 2	2 4		4 5	2 2		4 5	2 5
				4 7	2 4		3 2	2 0		4 7	3 0	F	4 1	—
							4 5	2 2		4 4	2 4		5 0	2 1
							4 2	2 2		5 0	2 2		5 0	2 4
							3 9	1 9		4 2	2 2		4 6	2 6
							3 9	1 9		4 0	2 3		4 6	2 0
							4 5	2 2		3 0	2 0		4 4	1 8
							4 9	3 2		4 4	2 1		5 3	2 8
										3 8	1 8		4 5	2 1
										4 5	2 5		4 0	2 0
										4 3	2 3		4 0	2 2
										4 2	2 1			
										4 6	2 7			
										3 9	2 4			
										3 8	2 2			
Total	5			18			24			32			27	

TABLE I—*Continued.*

TRANSVERSE DIAMETER OF NASOPHARYNX AND INTERTUBAL DISTANCES

7–8			8–9			9–10			10–11			11–12			12–13		
Sex	Tv. Diameter	Intertubal distance	Sex	Tv. Diameter	Intertubal distauce	Sex	Tv. Diameter	Intertubal distance	Sex	Tv. diameter	Intertubal distance	Sex	Tv. diameter	Intertubal distance	Sex	Tv. diameter	Intertubal distance
M.	4·8	2·5	F.	5·3	2·5	M.	5·1	2·0	M.	5·2	3·2	M.	3·8	2·2	F.	4·8	2·2
	4·8	2·6		5·0	2·3	F.	4·5	2·0		3·8	2·6		5·2	3·4		4·5	2·5
	5·2	2·3		5·2	2·3					4·8	2·5					4·2	2·4
	4·9	2·5		4·0	2·5				F.	5·2	3·2						
	5·0	2·5		4·5	2·1					5·7	2·4						
F.	5·2	2·2		4·2	2·0												
	5·3	2·8															
Total	7			6			2			5			2			3	

TRANSVERSE DIAMETER OF NASOPHARYNX AND INTERTUBAL DISTANCES. IN AGE GROUPS.

15–16			17–18			19–20			20–21			21–22		
Sex	Tv. diameter	Intertubal distance	Sex	Tv. diameter	Intertubal distance	Sex	Tv. diameter	Intertubal distance	Sex	Tv. diameter	Intertubal distance	Sex	Interdiameter	Intertubal distance
M.	5·0	4·0	F. F.	5·6 5·3	2·3 2·9	F.	5·5	3·2	F.	5·2	3·9	F.	4·6	2·6
22–23			24–25			29–30			38–39			41–42		
M.	6·0	2·8	F.	5·4	2·0	F.	5·1	2·8	M.	5·7	2·8	F.	5·8	3·2

Muscular Contraction in the Nasopharynx

As the survey proceeded, a considerable degree of contraction of the inter recess diameter was occasionally observed when patients were in light phases of anæsthesia with the gauge in position, and on one occasion, while the patient was retching, the examining finger was tightly gripped between the orifices of the eustachian tubes moving medially

Such extensive muscular contractions in the upper part of the nasopharynx and its anterior wall in relation to the cartilaginous eustachian tube, came as a surprise, as traditionally these areas are regarded as relatively rigid structures Its mobility under anæsthesia suggested that its power of contraction might have an important physiological function and to obtain further information on this, the programme of recordings was continued

Movement in this area has now been frequently demonstrated during light phases of anæsthesia and most often when the anæsthetic sequence is pentothal gas and oxygen, ether and trilene When it is present the inter recess diameter contracts during inspiration and expands with expiration with a range of the order of 0 5 cm Should gagging occur the degree of contraction is much greater and under these conditions the inter recess diameter may diminish by 2 cm or more

TABLE II

CONTRACTION

Sex	Age	Inter recess diameter cm	Inter recess contraction cm	Inter tubal distance cm	Inter tubal contraction cm
M	5	4 3	0 5	2 2	—
M	7	5 2	0 5	2 3	—
F	7	5 3	0 5	2 8	—
F	7	5 6	0 5	3 0	—
F	8	4 5	0 3	2 1	—
M	10	5 2	1 3	3 2	—
F	10	5 2	2 0	3 2	—
M	11	5 2	—	3 4	1 6
F	12	4 8	1 0	2 2	—
F	12	4 8	0 6	2 4	—
F	17	5 6	1 6	2 3	—
F	20	5 2	—	3 9	0 7
F	21	4 6	0 4	2 6	—
F	41	5 8	2 8	3 2	—

Contractions of the intertubal distance have only been measured on two occasions One in a boy aged 11 whose inter recess diameter was 5 2 cm and intertubal distance 3 4 cm The latter contracted to 1 8 cm indicating a medial movement of each eustachian tube of 8 mm The second was in a girl aged 20, with an inter recess diameter of 5 2 cm The intertubal distance was 3 9 cm and this contracted to 3 2 cm

Comment

1. During the recording of nasopharyngeal measurements three varieties of anatomical structure have been recognized digitally in the antero-lateral wall of the nasopharynx in the region of the pharyngeal eustachian tube.

(*a*) In the first of these the area has an even surface with absence of tubal prominence, and the introitus to the eustachian tube is recessed. This type is usual before the age of 6 years, though it may also occur in older children.

(*b*) In the second group there is a well defined rounded prominence of the nasopharyngeal part of the eustachian tube and the posterior lip of the tubal entrance is also prominent. After the age of 6 this is the usual finding, but it may be present in much younger children.

(*c*) The third group is only occasionally encountered and comprises less than 5 per cent. of cases. It presents as a foreshortening of the pharyngeal part of the eustachian tube, and the prominence beneath the antero-lateral wall of the nasopharynx is ill-defined. It does not appear to be pathological nor associated with tubo-tympanic dysfunction. When it is present the distance separating the tubal openings is of the order of 3·4 cm.

TABLE III.

THE LATERAL DIAMETER OF THE NASOPHARYNX IN 142 PATIENTS BETWEEN 2 AND 42 YEARS IN RELAXATION UNDER GENERAL ANÆSTHESIA.

Age Groups in years	Average	Transverse diameters in cm.	
		Maximum	Minimum
	cm.	cm.	cm.
2–3	4·06	4·3	3·7
3–4	4·2	4·8	3·2
4–5	4·3	5·2	3·2
5–6	4·5	5·4	3·8
6–7	4·7	5·7	4·0
7–8	5·0	5·3	4·8
8–9	4·7	5·3	4·0
9–10	4·8	5·1	4·5
10–13	5·1	5·7	3·8
15–20	5·3	5·6	5·0
20–30	5·3	6·0	4·6
38–40	5·75	5·8	5·7

2. In view of the wide range in measurements in each age group and the variations in anatomical structure in the antero-lateral wall of the nasopharynx, the calculation of tissue r dosage from the radium applicator

requires an individual measurement of the space in cases of tubal dysfunction

3 The presence of extensive muscle contractions in the upper nasopharynx in light anæsthesias suggests physiological muscular activity in this region in respiration, deglutition, phonation and singing, and would indicate that local inflammatory reactions, adenoid hypertrophies and reticuloses will impede this activity

Summary

The transverse diameter of the nasopharynx and the distances separating the eustachian tube orifices have been measured under anæsthesia in patients between the ages of 2 and 40 years The dimensions increase with age in a linear graph up to the age of 8 years

During the survey contractions in the transverse diameters have been observed from voluntary muscle action and it is suggested that under normal physiological conditions there is a continuous muscle movement in the upper nasopharynx It would seem probable that this produces a rhythmic and also intermittent pressures on the adenoid area in children, and that under pathological conditions this physiological action is impeded

Acknowledgments

I would like to thank Mrs Harris of the Medical Secretarial Staff at the Children's Hospital, Birmingham, and Miss Hadley, Ward Secretary at the Queen Elizabeth Hospital O R L Department, for their help during the progress of the investigation, and also Mr Dee, Medical Photographer in the Queen Elizabeth Hospital for the photograph

REFERENCE

REMAGGI P L (1955) *l Oto Rino-Laring Ital*, **35**, 231

AUDITORY TRAINING AND SPEECH AUDIOMETRY

By R. H. HUNT WILLIAMS (Reading) and DANIEL LING (Reading)

THE rapid development of hearing aids over the past few years had led to a wide interest in the use of residual hearing. Much of this interest has been focused on deaf and partially deaf children. It is well known that hearing loss, if present during childhood, may prevent the development of speech (Morley, 1957).

Conductive deafness (i.e. impairment centred in the outer or middle ear) can be overcome and the patient's hearing for speech brought to normal by the use of a hearing aid. Perceptive deafness (i.e. impairment centred in or beyond the cochlea) however slight as measured by pure tones, may prevent words being heard correctly, whatever the intensity to which they are amplified (Davis, 1948; Silverman, 1950, Huizing and Reyntjes, 1952; Meyerson, 1956). These facts are clearly illustrated by the use of speech audiometry. For this type of audiometry standardized records of speech are played, usually through earphones, and the percentage of words heard correctly at any given intensity is scored on a chart known as a speech audiogram.

In the present study, the effect of auditory training on five children with perceptive deafness, of various ages from 6-10 years, was observed, and progress measured with speech audiometry. In no case is the full medical ætiology available. A comparison between monaural and binaural hearing for speech was also made at the end of the study. Pure tone loss is given in each case at 250, 500, 1,000, 2,000 and 4,000 cycles per second. With the exception of Case CXI, these figures were constant to within 5 decibels.

Literature on the subject of auditory training is scanty. Brown (1949) has reported improvement in hearing for speech in adults with moderate hearing loss. Di Carlo (1948) gives data on hard of hearing veterans. Hirsch (1952) states that it is unusual for auditory training to change hearing loss for speech.

The writers could find no record of any controlled experiment in which the hearing for speech of school-age children was assessed by speech audiometry before and after auditory training, in which lip reading was suppressed. Experiments in which speech perception was measured by combined lip reading and hearing have, however, been carried out by Hudgins

(1953) in America and Clarke (1953) in England Wedenberg's (1951) work in Sweden, which was concerned with hearing only, was confined to children of pre-school age

The Training

The training was carried out in an experimental class, which was formed by the Reading Education Authority in 1955, for partially deaf children The classroom is equipped with a Westrex group hearing aid, the output from which may be controlled to within 5 decibels up to a maximum of 115 decibels The auditory training programme consisted of normal class lessons and stories recorded at approximately constant levels of intensity In addition, definite practice was given in listening to a wide variety of speech and other sounds The group aid was used during the course of the study for an average of 20 hours weekly

The Tests

Pure tone audiometry was carried out with a Maico audiometer The speech audiometry was designed and recorded by one of us (D L) using monosyllabic words and methods suggested by Mr Michael Reed * The words were grouped in lists of ten all of which were illustrated by appropriate pictures, to which each child was required to point when tested at various known intensity levels Each group of ten words was subdivided into two parts containing five similar vowel sounds, e g , "man", "pan", "van", "lamb", "hand" Thus, the test was essentially of the discrimination of consonants The recording, a permanent disc, was made at the London studios of His Master's Voice, and was standardized on fifty ears of normal children These normal responses were averaged and showed 100 per cent recognition at 30 decibels

Method

Pure tone and speech audiograms (below) were obtained on three occasions on each child during the period of study, and each ear was subsequently trained at appropriate levels of intensity All cases initially were intolerant to sound intensity above certain levels, the points at which the initial curves cease By working just below these and increasing the intensity in 5 decibel steps over a period of months, all intolerance up to 115 decibels was overcome Binaural responses were measured at the end of training, using earphones paired in output

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* Psychologist Audiology Unit London

Data

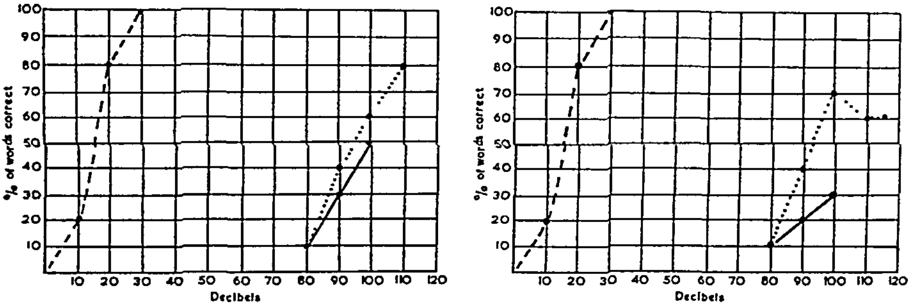
Case I

C.B., a congenitally deaf girl, aged 7 years, of normal intelligence, who had spent the past three years in a school for the deaf. She had worn a hearing aid for 4 years.

Pure Tone Loss

Left						Right				
250	500	1,000	2,000	4,000	c p s	250	500	1,000	2,000	4,000
65	85	95	90	95	Db loss	60	75	90	90	95

Speech Audiogram



----- Normal ——— March. December.

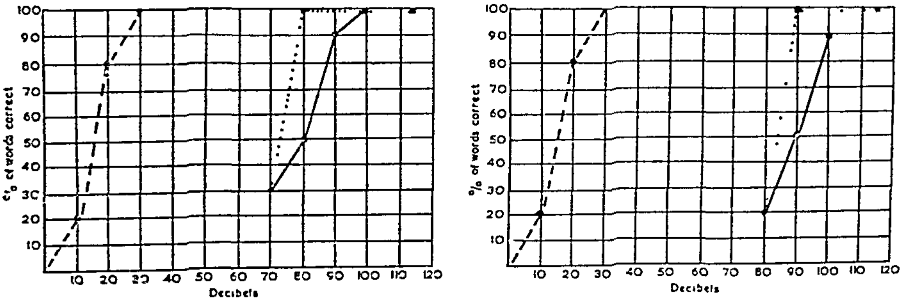
Case II

J.N., a congenitally deaf girl, aged 8 years, of normal intelligence, who had spent the past three years in normal schools and had received speech therapy for three years. During this period she had worn a hearing aid.

Pure Tone Loss

Left						Right				
250	500	1,000	2,000	4,000	c p s	250	500	1,000	2,000	4,000
30	35	55	85	85	Db loss	30	45	75	90	90

Speech Audiogram



---- Normal ——— March December

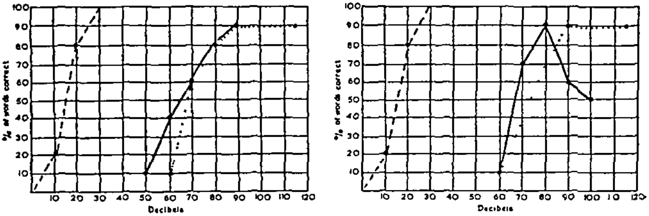
CASE III

M.O., a congenitally deaf girl, aged eight years, of superior intelligence, who had spent the past 3 years in normal school and had worn a hearing aid during this period.

PURE TONE LOSS

Left 250	500	1,000	2,000	4,000	c p s	Right 250	500	1,000	2,000	4,000
30	40	50	60	65	Db *March* Db	40	50	70	70	75
45	45	55	70	65	Db *December* Db	55	55	80	80	75

SPEECH AUDIOGRAM



---- Normal —— March December.

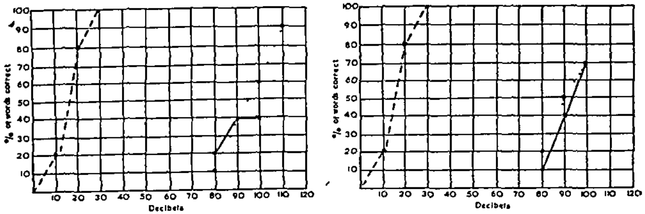
CASE IV

E.G., a congenitally deaf girl, aged 6 years, of superior intelligence, who had spent one year in a normal school and had received speech therapy for two years. During this period she had worn a hearing aid.

PURE TONE LOSS

Left 250	500	1,000	2,000	4,000	c p s	Right 250	500	1,000	2,000	4,000
40	55	50	65	40	Db loss	35	60	70	80	60

SPEECH AUDIOGRAM



---- Normal —— March. . December

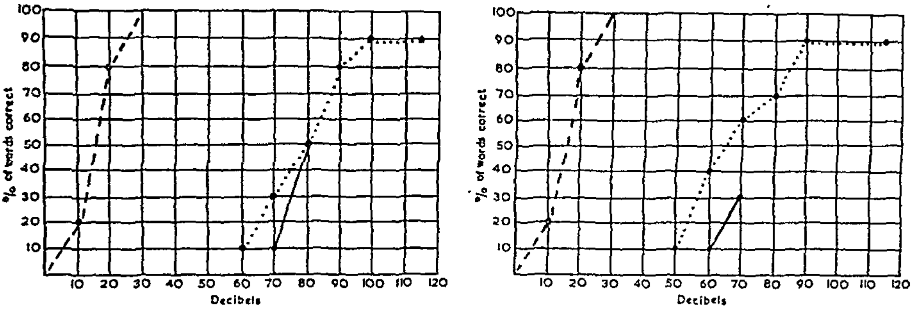
CASE V

A.S., a normally intelligent boy, aged 9 years, who had spent the past 5 years at special schools for the deaf. He had worn a hearing aid for 6 years. His deafness was acquired at 2 years of age.

PURE TONE LOSS OVER PERIOD OF STUDY CONSTANT TO WITHIN 5 DECIBELS.

Left						Right				
250	500	1,000	2,000	4,000	c.p.s.	250	500	1,000	2,000	4,000
40	60	70	80	90	Db. loss	30	50	70	75	90

SPEECH AUDIOGRAM.

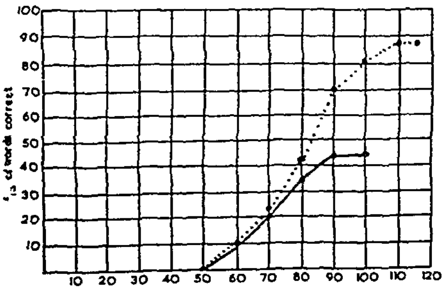


---- Normal. ——— March. December.

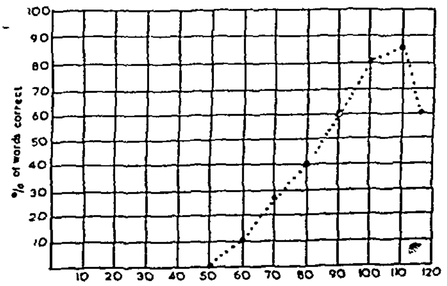
TABLE OF SCORES OVER INTENSITY RANGE 50–115 db.

Intensity Decibels	50	60	70	80	90	100	110	115
Initial worse ears	2	8	14	36	40	32		
Initial better ears		4	26	32	44	54		
Initial average	1	6	20	34	42	43		
Final worse ears		4	14	34	72	84	82	82
Final better ears	2	10	30	54	70	80	90	90
Final average	1	7	22	44	71	82	86	86
Binaural average	2	12	28	40	60	80	84	62

SUMMARY



Average of monaural scores.



Average of binaural scores.

In this study of a small series of cases carried out over a 9 month period results show that monaural speech discrimination was raised from an average maximum of 43 per cent to 86 per cent and that the range of intensities over which words could be recognized was considerably extended However, the intensity levels at which speech first became intelligible did not change significantly

Only in Case V was binaural hearing superior to that of monaural hearing in the better ear

Discussion

Under normal conditions the intensity of speech changes constantly and rapidly The ability to discriminate sound over as wide a range of intensity as possible is, therefore, essential to adequate hearing for speech In the above five cases the factors preventing adequate use of hearing were (i) high thresholds (deafness) (ii) poor discrimination, (iii) subjective distortion and (iv) intolerance

HIGH THRESHOLD

The use of a hearing aid alone merely raises sound to a level which can be perceived In the cases under review this was insufficient Hearing aids had been worn for a number of years yet after auditory training, use of residual hearing was improved

POOR DISCRIMINATION

In these cases of peripheral deafness poor discrimination was directly related to lack of auditory experience which adversely affected language development The role which language development plays in the discrimination of speech is discussed in detail by Fletcher (1929), Fry (1955) and Miller (1951) Briefly, it may be said that familiarity with language makes it possible for the listener to supply missing sounds and words

SUBJECTIVE DISTORTION

Subjective distortion may be defined as a decrease in perceptive ability resulting from an increase in intensity above a certain level This should not be confused with objective distortion which may arise from the amplification circuit Hudgins (1953) Subjective distortion has been related to the recruitment phenomenon by Huizing and others (1952) Case III shows the typical helmet shaped curve in the right ear The shape of this curve altered during the course of auditory training Subsequently a saturation level at 90 per cent was reached (This was surpassed after further training) To obtain this result recorded speech was played to the child at optimum level (80 db) until familiar The intensity was then gradually increased into the area where subjective distortion occurred

Subsequent studies carried out on a further group of six children, three of whom had this characteristic curve, responded similarly. The writers consider that this observation merits further study.

INTOLERANCE

The intensity at which sound became uncomfortable for the listener varied. In no case could sound be tolerated initially above 100 decibels. Case V illustrates the exceptional degree to which tolerance was raised with resulting improvement in articulation score.

Indirect results of this training were (*a*) an improvement in speech and (*b*) an improved ability to hear conversation with an individual hearing aid. (*c*) A remarkable feature was the spontaneous acquisition and use of new words by all of the children throughout the study. It is suggested that such improved use of residual hearing leads to a higher level of social adequacy (Davis, 1948).

Conclusions

In formulating an auditory training programme for the partially deaf and assessing the benefits derived therefrom, the use of speech audiometry is essential.

Acknowledgments

We are indebted to Mr. P. S. Taylor, Chief Education Officer for Reading, and to Miss Edwards, Headmistress of the George Palmer School, whose pupils co-operated in the standardization of the audiometric tests. Also to Mr. P. Gorman, Cambridge University, for his helpful criticisms.

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NERVE DEAFNESS ASSOCIATED WITH SARCOIDOSIS

By R E GRISTWOOD (Adelaide South Australia)

SARCOIDOSIS, or Besnier-Boeck-Schaumann disease, is a condition remarkable no less for its protean clinical manifestations than for the paucity of symptoms in relation to the extent of the lesions, which may be widely disseminated in the body Although any organ may be involved in the sarcoid lesion, lymph nodes, lungs and skin are the most commonly affected, and less often the uveal tract of the eye, and the bones of the extremities The pathological features of sarcoidosis are those of a low-grade granulomatous inflammatory process characterized by the presence in any organ or tissue of epithelioid-cell tubercles with absence of central caseation Multinucleated giant cells are a conspicuous component of many of the lesions The lesions may be widely disseminated, the distribution of the lesions varies greatly, and their effects depend upon the situation in which they occur and upon the size of the sarcoid clusters The clinical course is usually chronic with minimal constitutional symptoms There may be, in some cases, acute phases with fever and malaise, and there may be signs and symptoms referable to the tissues and organs involved

Involvement of the nervous system in cases of sarcoidosis is comparatively rare, occurring in 1 to 5 per cent of all cases (Hook, 1954) The incidence is somewhat higher if cases are taken into account which lack clinical manifestations but exhibit slight abnormalities in the cerebro spinal fluid The disease may involve any part of the nervous system Facial paralysis, usually transient and sometimes recurrent, is one of the most common signs, occurring in about 50 per cent of these cases (Colover, 1948) The central nervous system may develop meningitis or encephalomyelitis, or there may be localized infiltration by sarcoid lesions, predominantly in the basal structures of the brain These conditions may be manifested clinically by generalized or focal disturbances of cerebral function, e g diabetes insipidus, resulting from invasion of the pituitary gland, is not uncommon

Nerve deafness, arising as a manifestation of sarcoidosis, is an event of considerable rarity Apparently deafness has never been observed as a complication of sarcoidosis in childhood McGovern and Merritt (1956) found, in their search of world literature up to February, 1953, that more than 3,000 cases of sarcoidosis had been reported, and of these 104 were children of ages from 2 months to 15 years. The authors recorded a further nine children with sarcoidosis observed by themselves, bringing the total number of cases in children to 113. Reports of neurological features were uncommon, apart from VIIth nerve paresis in four cases.

The possibility of deafness occurring in sarcoidosis in adults is mentioned by Longcope and Freiman (1952), and articles by Bouchet and Debain (1947), Leitner (1949), and Essellier *et al.* (1951) quote briefly a few of the cases that have been reported in the literature. Colover (1948), in a survey of 118 cases of sarcoidosis involving the nervous system, remarks in his discussion on the frequency of cranial nerve lesions that "nerve deafness was noted in eight cases, and in four cases there was disturbance of the vestibular branch of the acoustic nerve".

In Table I are presented all those case histories of nerve deafness, with or without vestibular upset, in association with sarcoidosis recorded in the literature.

TABLE I

CASES OF SARCOIDOSIS ASSOCIATED WITH NERVE DEAFNESS

Author	Age and sex	Clinical features	Otological findings	Histology
MacBride (1923)	43 Female	Bilateral uveitis and conjunctivitis, and bilateral parotid swelling in association with a transient peripheral neuritis affecting the lower limbs The parotitis disappeared in one month Several months later facial paralysis with *slight deafness*, first on one side and then four months later on the other, appeared Cranial nerves V and X were also affected	Slight bilateral deafness (Detailed findings not recorded)	None available
Souter (1929)	40 Male	Uveitis and parotid swellings developed with mild fever, weakness and cough, facial paralysis, skin eruptions, joint pains and absence of deep reflexes in the lower limbs The iridocyclitis persisted, facial paralysis recovered, and the knee jerks returned, but he gradually deteriorated and died 7 months after the first examination *Autopsy* showed enlarged hilar glands, miliary tubercles throughout both lungs, and infiltration of the myocardium of the left ventricle by tuberculous granulation tissue	"right ear heard a light watch at 2in , and the left not on contact " There was a history of discharge from the left ear for a long period in childhood (Detailed findings not recorded)	*Sarcoidosis* Tuberculous granulation tissue without caseation (Tubercle bacilli were claimed to have been found in the heart muscle, by trituration)
Mohn (1933)	28 Female	For about 5 years had suffered from brief bouts of abdominal pain accompanied by nausea and vomiting Was admitted to hospital on account of one such attack with slight fever, when she developed bilateral facial paralysis and paresis of the soft palate, followed 3 weeks later by bilateral enlargement of the parotid and lacrimal and sub maxillary glands Five weeks after the onset of her facial paralysis, she developed *severe bilateral deafness*, flaccid paresis of arms and spastic paralysis of the legs Bilateral iridocyclitis appeared at this time, and chest X ray showed slight increase of hilar shadows and a small area of infiltration in the left upper lobe Signs and symptoms slowly diminished but 9 months after the onset, she still had facial paralysis and uveitis	Severe bilateral deafness (type not specified)	None available

Nerve Deafness Associated with Sarcoidosis

Author	Age and sex	Clinical features	Otological findings	Histology
Garland and Thomson (1934)	31 Female	Iridocyclitis and parotid swelling Severe generalized polyneuritis with R VII nerve paralysis and *progressive bilateral deafness*, which became almost *complete 8 months after the onset of* the illness Vision and hearing later improved, the facial palsy recovered, and the deep reflexes returned	*Bilateral nerve deafness* superimposed on a mild middle ear deafness (*result of* childhood otitis media)	*Sarcoidosis* (biop of skin lesion c front of leg)
Levin (1935) 1	28 Male	Bilateral uveitis and parotid swelling, transient L sided facial paralysis and diabetes insipidus At the time of recovery *of the facial paralysis deafness* and unsteadiness of gait appeared No vertigo Acuity of hearing decreased gradually over a period of several weeks and then remained stationary with persistent tinnitus Anosmia, sensory loss on the centre of face, and an inconstant extensor plantar response	Both drum heads normal Weber→ R A C > B C in both ears Audiometry showed *bilateral nerve deafness* with high tone loss The response of both labyrinths to rotation and caloric stimulation was decreased	None available
2	30 Male	[illegible] symptom	*Bilateral nerve deafness* and impaired labyrinthine function	None available
Zollinger and Meyenburg (1941)	21 Male	Headache and vomiting Generalized aches and pains Intermittent pyrexia, diplopia, ataxia, nystagmus and deafness Extensor plantar responses *Autopsy* confirmed sarcoidosis involving nervous system *C S F* cells 21 per c mm protein 46 mgm per cent abnormal colloidal gold curve	Deafness (no details given)	*Sarcoidosis* Diffus perivascular cuffin with epithelioi cells in brain stem cerebral hemispheres and cerebellum
Erickson, Odom and Stern (1942)	31 Male	*Patient had a sarcoid lesion on the* upper lip and developed the symptoms of increased intracranial pressure, deafness and tinnitus in the L ear, and recurring attacks of vertigo Both ears eventually showed slight loss of hearing Posterior fossa exploration revealed a chronic adhesive arachnoiditis with herniation of the cerebellar tonsils [illegible] brain-stem The nerve roots were free	Bilateral slight hearing loss	*Sarcoidosis* Pia arachnoid was infiltrated by nodule consisting of roun cells, plasma cells epithelioid cells an giant cells Similar cell collections were presen around small blood vessels *in* the substance of the pon and cerebellum
Ernsting and Sillevis Smitt (1944)	31 Female	In November 1937 the patient developed persistent frontal headaches, listlessness and anorexia, with weight loss, and had occasional nausea and vomiting with dizziness In March, 1938, bilateral iridocyclitis appeared Chest X ray showed pro [illegible]	Drumheads normal Auditory acuity reduced to whisper at 3 metres left and 10 cm on right Rinne bilaterally positive Weber lateralised to left Both labyrinths react normally to caloric tests	None available

Author	Age and sex	Clinical features	Otological findings	Histology
Hedges and Taylor (1952)	21 Female	The patient, a negro woman who had previously enjoyed good health, developed general malaise, joint pains and dyspnoea on exertion in March, 1951 In May, 1951, iridocyclitis with "mutton fat" KP and iris nodules appeared In July she had headaches, tinnitus, vertigo and bilateral deafness Chest X ray showed prominent hilar gland enlargement, and biopsy of a small supraclavicular node confirmed the diagnosis of sarcoidosis	Drumheads normal Audiometry showed a *bilateral low tone nerve deafness of* 40 db loss which gradually improved over a period of 9 months to a 30 db loss for the low frequencies Rotation and caloric tests revealed hypo function of the vestibular labyrinths After 6 months, the right labyrinth had almost returned to normal while the left showed complete loss of function	*Sarcoidosis* (biopsy of supraclavicular node)
Hook (1954) 1	36 Female	Transient attacks of bilateral facial paresis accompanied by swelling of parotid glands and agenesia in 1939 In 1940 uveitis developed with recurrence of parotitis and facial paresis Chest X ray showed bilateral hilar gland enlargement and fine mottling throughout both lungs Mantoux negative 1941 46 symptom free In 1947 she had frequent headaches, transient facial paresis, amenorrhoea, polydipsia and polyuria, with a return of uveitis accompanied by *bilateral deafness* and *rotary vertigo* All symptoms disappeared completely in a few months	*Bilateral nerve deafness*	None available
2.	63 Female	In 1944 had bilateral iridocyclitis followed by *dizziness* and gait disorders lasting a few months In 1945 she again experienced dizziness and gait disturbances, accompanied by profound right sided deafness Some time later, hearing improved, but she was troubled by tinnitus Easily fatigued and had periodic sleep disturbances Transient R facial paralysis Chest X ray showed scattered densities throughout both lungs Mantoux negative In 1952 patient was still troubled with tinnitus and occasional mild vertigo, but was able to manage unaided	*Bilateral deafness* R > L *of nerve type* with sluggish caloric reactions	None available

Recently I had the opportunity of examining a case similar to those already reported, the details of whose history, investigation and progress are given below.

History

A woman, aged 26 years, single, and the mother of a 10 month old boy, was referred on 17.5.57 to the Ear, Nose and Throat Clinic in Galashiels on account of bilateral deafness of sudden onset which had persisted for 6 weeks. At the beginning of March, 1957, one month prior to the onset of deafness, she developed congestion and soreness of both eyes, a condition which had endured and was associated with gradual impairment of distant vision. In the middle of March, a right-sided facial palsy appeared, and she developed ataxia with a tendency to fall to the right. At the end of March the ataxia became less noticeable, the facial paralysis recovered completely, and three days following that she awakened one morning with severe bilateral deafness She had previously had normal hearing with no history of ear trouble The deafness had remained constant in degree and was associated with a "clicking" tinnitus in the right ear related to jaw movement In addition she had lost about 19 lb in weight during the preceding 2 months, and the complexion of the face and neck had become darker She had some loss of appetite, and was easily fatigued She had no respiratory symptoms and no past history of any illness apart from mumps and measles in childhood Her illegitimate son was thriving well There was no family history of tuberculosis She was admitted to the Eastern General Hospital, Edinburgh, on 22 5 57 for fuller investigation, as the initial examination had revealed bilateral nerve deafness and iritis

Examination

The patient was a rather thin young woman who had a dusky complexion of her face, neck and hands There was a well-marked irregular patchy dark-brown pigmentation of the mucosa in the floor of the mouth, and on the buccal surfaces of the upper and lower lips and gums There was no pigmentation elsewhere on the body and no skin lesions

Clinical examination of the chest was negative, there was no enlargement of liver or spleen and no glandular masses One small firm lymph node, the size of a pea, was palpable in the right supraclavicular region between the two heads of origin of the sternomastoid muscle This node was later excised for biopsy

Apart from a tendency to stagger slightly to the right on walking along a straight line, neurological examination was negative The right-sided facial paralysis had completely recovered and there was no nystagmus

The cardiovascular system was normal B P was 110/60 and there was no abnormality in the urine

Eyes Both eyes showed ciliary flush and irregularity of the pupils which reacted to light Instillation of 1 per cent atropine sulphate caused disappearance of the discomfort and ciliary flush within a few days The ophthalmologist reported 2 weeks later—"Both eyes white, no ciliary or conjunctival flush both pupils irregular and show multiple posterior synechiae, with deposit of pigment over the anterior surface of the lens, indicating old iritis, no fresh K P seen

' Fundus right eye details not very clearly seen because of exudate over the lens, but no gross pathology seen

"Fundus left eye media clear, disc normal, blood vessels show definite thickening of arteries with nipping of veins at arteriovenous crossings, suggesting early setting of arteriosclerosis

"No tubercle seen either in the iris or the fundus "

Nose, Pharynx and Larynx Normal

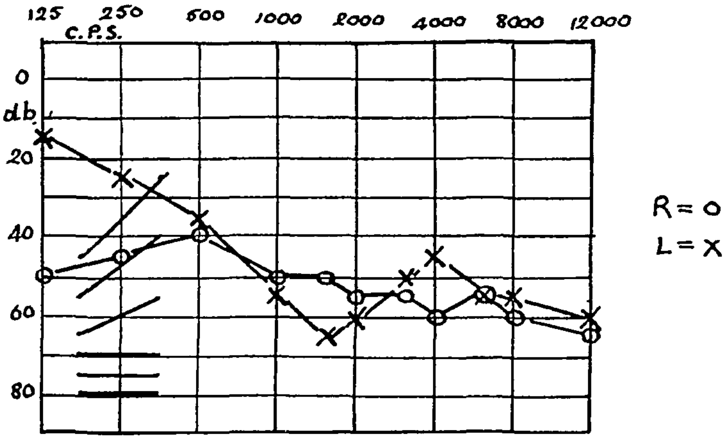
Otological Investigation

Both tympanic membranes were normal. The eustachian tubes were patent.

Cochlear Function:

		Right ear	Left ear
Whispered voice (normal 30 ft.) ..		6 in.	6 in.
Conversational voice (normal 30 ft.) ..		1½ ft.	3 ft.
Weber		lateralized	to left
Rinne	256	+ve.	+ve.
	512	+ve.	+ve.
	1,024	+ve.	+ve.
	2,048	+ve.	+ve.
Absolute bone conduction		reduced	reduced

Audiogram 23.5.57.



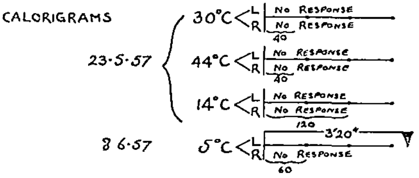
The loudness recruitment phenomenon was present in the right ear at 250 c.p.s.

Vestibular Functions. Stance with eyes closed normal. Tendency to walk to right side with eyes closed.

No spontaneous nystagmus was present with the head erect.

No past-pointing, dysdiadokokinesis, inco-ordination, or positional nystagmus.

Caloric responses were absent on the right side and severely impaired on the left.



Radiological Investigation

Chest Diffuse faint nodulation throughout both lungs associated with several larger rounded opacities and bilateral hilar gland enlargement (see Fig. 1).

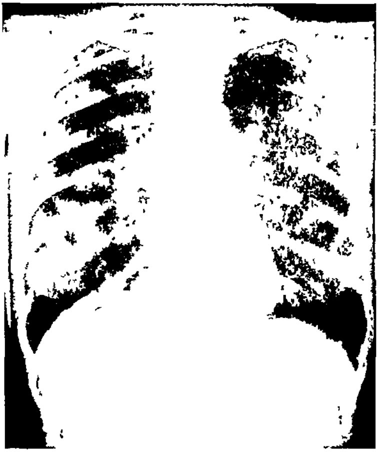


FIG 1

Radiograph of chest showing hilar glandular enlargement, and sarcoid infiltration in both lungs

Tomograms. Paratracheal and bilateral hilar glandular enlargement. Numerous rounded lesions varying from 1 to 3 cm. in diameter throughout both lung fields.

Radiographs of the skull in all positions, of the petrous bones, of the sinuses, and of the hands and feet showed no abnormality.

Special Investigations

(1) *C.S.F.* Crystal clear and colourless. Resting pressure 100 mm. with free rise and fall on jugular compression.

Cells = 6 per c. mm.
Protein = 55 mgm. per cent.
Sugar = 48 mgm. per cent.
Chloride = 724 mgm. per cent.
Colloidal gold curve = 222,221 0000
W.R. negative.

(2) *Blood.*

Hæmoglobin = 13·2 g. per cent.
White cells = 6,000/c. mm. Normal differential count.
E.S.R. = 30 mm. in the first hour.

Plasma proteins	Albumin	3·65	g. per cent.
	globulin	3·25	
	Total	6·90	
Serum electrolytes	Ca^{++}	11·8	mgm. per cent.
	Na^{+}	310	
	Cl^{-}	546	
	K^{+}	19·4	

Blood sugar 103 mgm. per cent.
Plasma cholesterol 208 mgm. per cent.
Wassermann reaction: negative.
Gonococcal complement fixation test: negative.

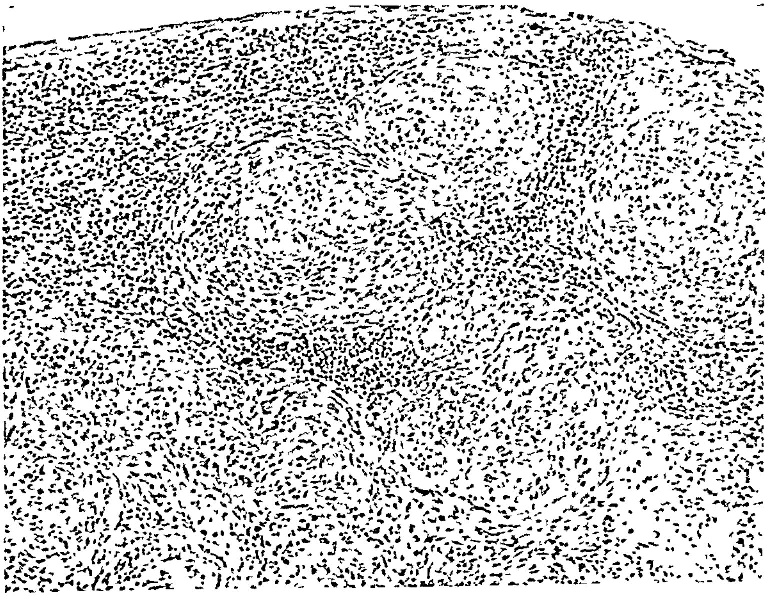


FIG 2.

Photomicrograph of lymph node (× 95) showing closely packed groups of epithelioid bodies containing a few multinucleated giant cells Note the absence of central necrosis

(3) *Mantoux Reaction* negative to 1/10,000, and 1/1000 positive to 1/100

(4) *Biopsy* of the supraclavicular lymph gland confirmed the diagnosis of sarcoidosis Fig 2 shows the closely-packed groups of epithelioid bodies containing multinucleated giant cells and the absence of central caseation

Progress

The patient remained completely afebrile and felt well during her stay in hospital The iritis rapidly settled on atropine instillations, the ataxia disappeared completely within a few weeks, and the deafness showed slight improvement to a conversational voice at 3 ft with the right ear, and 5 ft with the left ear, after three weeks

On 24 6 57 she was transferred to the care of Dr Ian W B Grant at the Northern General Hospital Edinburgh for treatment with a combination of prednisolone and anti-tuberculous chemotherapy For one month she received prednisolone 20 mgm daily with Pycamisan P H 10, 4 cachets b d , after which the dose of prednisolone was reduced to 15 mgm daily for a further 2½ months Thereafter the amount of prednisolone was further diminished to 10 mgm daily on which dosage she is at present (November, 1957) being maintained In order to preserve adrenal function while receiving prednisolone the patient received an intra-muscular injection of ACTH gel 30 units fortnightly After 2 weeks of prednisolone therapy the hearing had improved to a conversational voice at 15 ft right and 12 ft left (8 7 57) and the caloric response of the left ear was more sensitive

By 23 7 57, after one month of therapy, her hearing had increased to a conversational voice at 25 ft right, and 15 ft left Caloric responses were now obtainable from both ears by a stimulus at 20° C for 1½ minutes

Since 23 7 57 there has been further gain in hearing clinically, and audiometrically there has been some 10 to 20 decibels of improvement for certain frequencies in both ears The hearing, however, is not yet normal There is a pronounced dip in the audiogram of the left ear between 1,000 and 4,000 c p s The chart of the right ear shows a 20 decibel loss for frequencies less than 2 000 c p s Both ears show a high tone loss The binaural loudness recruitment phenomenon has persisted throughout the whole period of observation Further improvement in the caloric responses has occurred, especially in the left ear, but both labyrinths are as yet unresponsive to irrigation with water at 44° C for 40 seconds The audiometric gains and caloric test changes at varying intervals during treatment are shown in the diagram, Fig 3

By 3 10 57 there was practically no trace of pigmentation on the gums or floor of the mouth She had put on weight, and there was pronounced mooning ' of the face

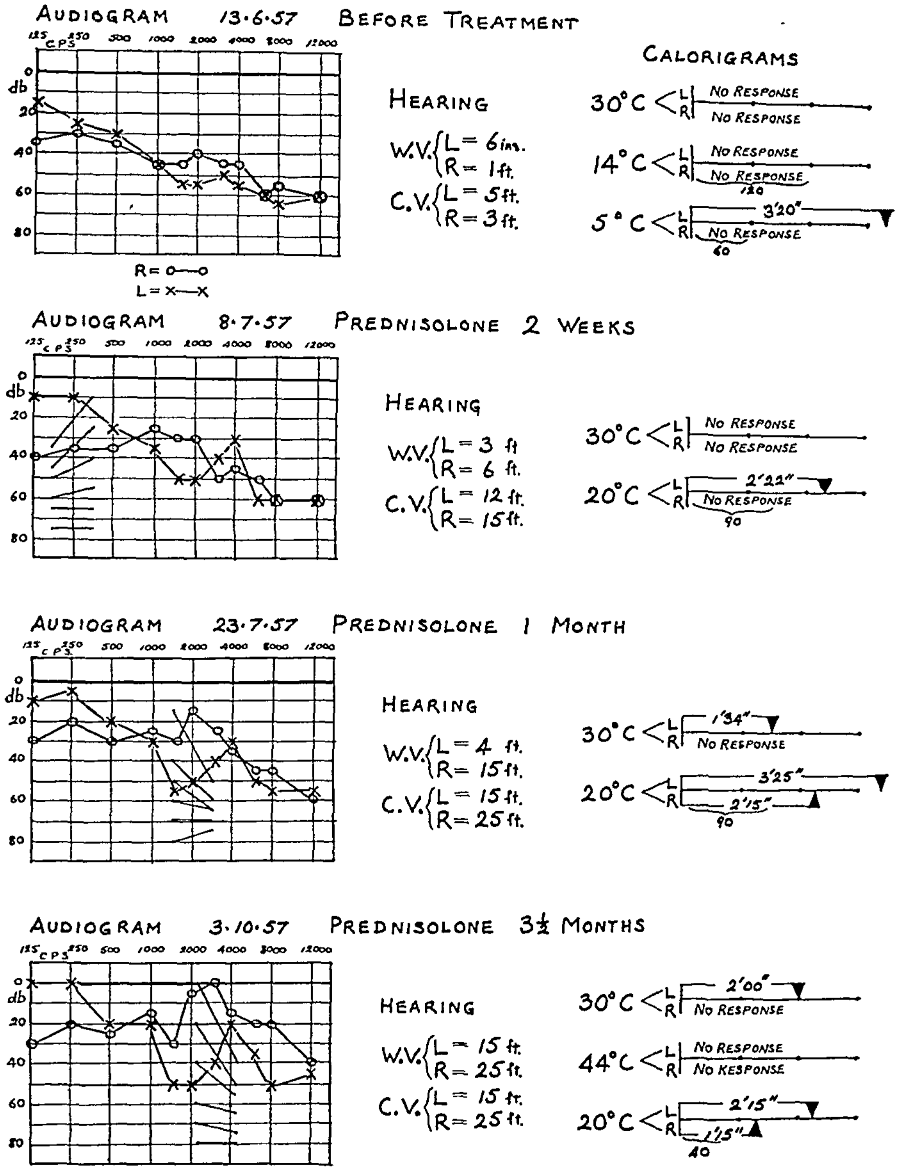


FIG. 3.

Audiograms, loudness recruitment phenomena and calorigrams at various stages of progress.

Discussion

The present case brings the total number of cases of deafness associated with sarcoidosis described in the literature to 13. In eight of these, two males and six females, the deafness has been confirmed as being inner-ear in type (Garland and Thomson; Levin (2); Ernsting and Sillevis-Smitt;

Hedges and Taylor; Hook (2); Gristwood). Absence of otological examination in the remaining five, three males and two females, leaves the diagnosis in doubt, although the fact that the co-existence of vestibular symptoms was reported in two (Zollinger and Meyenberg, Erickson *et al.*) is strongly suggestive that the VIIIth cranial nerve was affected.

One case of middle-ear deafness with sarcoidosis has been described by Colover, but is not considered in this review which is confined to patients with nerve deafness.

The high incidence of uveitis, which occurred in 11 (84·6 per cent.) of the patients in the present series is a notable feature, which contrasts with Colover's report that 56 per cent. of patients with sarcoidosis of the nervous system had involvement of the uveal tract.

Facial paralysis occurred in eight of the present cases, an incidence of 61·5 per cent. In three, the paralysis was bilateral; in five, unilateral. With the exception of two patients with unilateral facial palsy, the remaining six had bilateral parotid gland enlargement at some stage in their illness. The parotid swelling preceded the onset of facial paralysis in two cases, and occurred concurrently in three instances; in one patient facial paralysis developed three weeks before parotitis appeared.

Vestibular upset had the same incidence in this series as facial paralysis, occurring in eight instances. Had caloric tests been carried out routinely on all these patients, it is probable that the incidence of vestibular abnormality would be even higher. Of the six patients on whom caloric tests were performed, only one case (Ernsting and Sillevis-Smitt) is recorded as having normal reactions.

Parotid swelling occurred in six cases (46·2 per cent.), all of whom had facial paralysis as noted previously. Sixty-two per cent. of Colover's series had parotitis.

Eight patients in the present group had sarcoid lesions in the lungs or hilar glands.

The cause of the nerve deafness in the cases of sarcoidosis reviewed is a matter for conjecture, and in the absence of autopsy material the problem must await a definitive solution. The hearing loss may be attributed to:

(1) *Toxæmia* affecting the organ of Corti or the cells of the spiral ganglion. To those who believe that the phenomenon of loudness recruitment is due to disease of the hair-cells in the organ of Corti, toxæmia as the cause of deafness deserves consideration, in view of the complete recruitment which persisted in my case throughout the period of observation.

(2) *Mechanical pressure on the auditory nerve* by multiple sarcoid deposits in the meninges.

(3) *Direct invasion of the nerve* itself by sarcoid with disruption of nerve fibres.

(4) *Infiltration of the brain-stem* by perivascular collections of sarcoid which interfere with conduction in the central auditory pathways.

Any or all of the above mechanisms may play a part in producing nerve deafness in sarcoidosis.

In a similar way the impaired caloric reactions reported in several cases may be due to sarcoid deposits infiltrating the vestibular branch of the acoustic nerve, or its central connections in the brain-stem, or to a toxic effect on the ganglion.

Following the introduction of ACTH and cortisone in the therapy of sarcoidosis, certain cases have shown dramatic improvement symptomatically and in the clearing of the pulmonary infiltration (Longcope and Freiman, 1952).

Since starting treatment with prednisolone and anti-tuberculous chemotherapy, my patient has shown progressive improvement in her deafness, and her hearing at the present time (October, 1957) has almost returned to normal. The caloric reactions which were initially absent in both ears, have returned in large degree in the left ear, but the right vestibular labyrinth has still a severe depression of its function.

Radiologically there has been a considerable improvement both in the glandular and the pulmonary lesions. This improvement is most probably related to the resolution of the sarcoid lesions in the presence of circulating prednisolone.

The pigmentation—a feature which has been difficult to explain especially as there was nothing to suggest sarcoid deposits in the adrenal glands—has become less marked since her condition began to improve, and the buccal pigment has disappeared.

Summary

1. A case of sarcoidosis confirmed histologically, with nerve deafness, ataxia, impaired vestibular function, transient facial palsy, uveitis, and pulmonary involvement is described.

2. The improvement in symptoms that occurred following treatment with prednisolone is recorded.

3. Twelve cases of nerve deafness complicating sarcoidosis are summarized from the literature.

4. The pathogenesis of the deafness in these cases is discussed.

Acknowledgments

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CLINICAL RECORDS

CHOLESTEATOMA CYST OF THE NECK

By PHILIP READING (London)

IN *October, 1953*, a male patient aged 54 presented himself at the Out-Patient Clinic complaining of purulent discharge from the left ear, and of an intermittent, painful swelling below and behind the ear.

He had suffered from chronic otitis of the left ear since childhood and had undergone several operations on it, but the aural discharge had continued. On two previous occasions an inflammatory swelling had appeared behind the mastoid process and had been incised with temporary relief. He was otherwise in good health.

At his first attendance a brawny swelling was found lying under the upper end of the sternomastoid muscle, and the attendant œdema spread over the angle of the mandible and the back of the head. The abscess was incised and a quantity of foul, thick matter evacuated. Culture of the pus gave a heavy growth of *Streptococcus fæcalis, Pyocyaneus* and *B. coli*. Recovery was rapid, but on a routine follow-up attendance three weeks later, an elastic swelling, the size and shape of a hen's egg, could be felt under the anterior border of the upper end of the sternomastoid muscle. The tumour was painless, and it could be moved backwards and forwards to a small extent; it appeared to be attached to the deep surface of the muscle, probably as a result of the previous suppuration. Trans-illumination and fluctuation could not be elicited, but it was considered that the bulk of the overlying muscle might have played some part in obscuring these physical signs.

A tentative diagnosis of branchial cyst was made; and the neck was explored through a transverse, crease-line incision. A cyst was found deep to the anterior border of the sternomastoid muscle; during the dissection its wall was ruptured, and a quantity of thick, grey pultaceous matter was aspirated. An extensive dissection, made rather difficult by adhesions from previous infection, enabled the cyst walls to be excised. Some sort of fibrous track was found (or unwittingly manufactured by the surgeon's manipulations) which led upwards and inwards between the fork of the internal and external carotid arteries. This was followed towards the base of the skull, where it petered out. Subsequent reflection suggests that this was nothing more than a fibrous strand, perhaps a condensation of the deep cervical fascia. Histological section of the cyst wall (Fig. 1) showed a squamous, stratified epithelial lining to be present.

Immediate progress was satisfactory, but within six months, the patient returned to the clinic, again complaining of swelling and pain in the same place. Again an inflammatory mass was to be felt, which, on incision discharged an

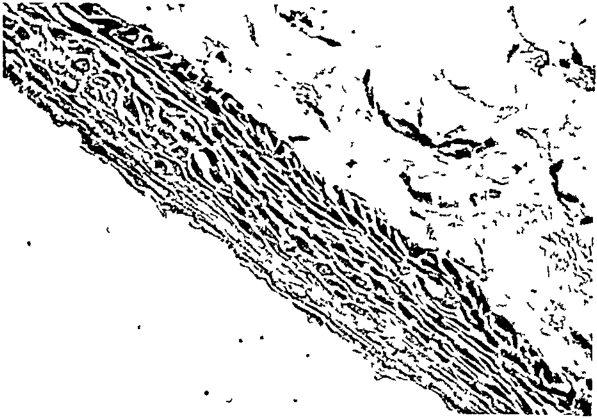

FIG 1

Lining of stratified squamous epithelium of cyst from Case I

ounce or so of thick offensive material Again, after the acute inflammatory reaction had subsided, a tense, elastic tumour was to be felt under the anterior border of the sternomastoid muscle

Luckily for the author, and incidentally for the patient who was becoming restive under these repeated and fruitless operations, a case report was published in 1955 by E Turnheim recounting a similar case history The patient had previously had a radical mastoidectomy, and had been troubled by recurrent swelling behind and below the mastoid process Incision and partial excision of the cyst had resulted only in its speedy recurrence Turnheim by an accurate dissection of the cyst walls was able to show that the neck of the cyst arose from an unopened air cell in the mastoid tip The radical removal of cyst wall, track and diseased air cell produced a complete and lasting cure

The cyst contained cholesteatoma, and the cyst wall was of stratified squamous epithelium Turnheim coined the name "Cholesteatoma Cyst of Neck", and he expressed the opinion that the cyst resulted from the outgrowth of cholesteatoma membrane from the unopened cell into the soft tissues of the neck

Armed with this knowledge, the author persuaded his somewhat reluctant patient to undergo yet one more operation The neck was re-opened, the cyst, which had grown again to the size of an egg, was dissected out of its bed, and a definite prolongation of its lumen could now be found to run upwards to end beneath the post-aural incision of the previous radical mastoidectomy A fine track was *found to lead from the interior of the cyst into the actual* radical mastoidectomy Excision of this track, together with the cyst, put an abrupt end to any further abscesses or swellings of the neck, and, somewhat surprisingly, it also stopped the slight foul otorrhœa which had been present from the mastoidectomy cavity over many years.

This case shortly was followed by another, very similar. A woman of 44, attended the Out-Patient Clinic in *October, 1957*, complaining of persistent discharge from the left ear and of intermittent, painful swellings below and behind the ear.

At the age of 5, she had undergone a left cortical mastoidectomy; at 12 she again entered hospital for the performance of a radical mastoidectomy upon the same ear. At the age of 30, she developed a post-aural abscess; and over the next years, she had several similar attacks of inflammation, but they had been repulsed by local application of heat and by the administration of sulphonamides and antibiotics.

The persistence of the aural discharge and the recurrence of the post-aural infection led her to submit to a revision of the radical mastoidectomy. The operating surgeon re-opened the post-aural incision, and he found that a small sinus which had been present at its lower end led into a mastoid cavity filled with cholesteatoma.

The post-operative course was disappointing; some granulations developed over the promontory and the ear did not become entirely dry. The incision healed, but the tissues beneath it remained indurated and tender.

Six years later she was re-admitted to hospital suffering from a brawny, inflammatory mass below and behind the ear. The swelling was needled, no fluid could be withdrawn. The infection subsided under anti-biotic therapy, but there still remained some thickening under the tip of the mastoid process. No definite tumour could be felt, but all the soft tissues in this region were thickened to the touch.

Once the treatment was stopped, there was an immediate recurrence of inflammation and the neck again became widely indurated. This settled after the administration of a second course of the same antibiotic.

On her admission in *1957*, a tense, elastic swelling could be felt under the mastoid tip; it appeared to be either under the sternomastoid muscle or within its substance. In the light of our previous experience and of Turnheim's paper, confident in our diagnosis of Cholesteatoma Cyst of the Neck, we proceeded to operation. An abscess cavity containing foul pus and cholesteatoma was found within the substance of the upper end of the sternomastoid muscle; the walls of the cavity were so tough and well defined that it was possible to strip them from the inflamed adjacent tissues. A definite neck was found to the cyst, leading upwards and opening into the floor of the mastoid cavity, apparently into the innermost part of the external auditory meatus. This was removed complete with the cyst, and the neck wound was sutured. Primary healing took place, and the patient has remained free till the time of writing from both cervical inflammation and from otorrhœa. Histological section of the cyst confirmed the presence of cholesteatoma and of a squamous, epithelial lining.

Turnheim's case appears to have originated in the escape of cholesteatomatous matrix from an air cell in the mastoid tip. Of the two reported in this paper, the first cyst appears to have come likewise from an escape of *matrix* from the msatoid cavity itself, and the other from some dehiscence in the deeper part of the floor of the external auditory meatus The presence of the cyst neck, connecting with the mastoid cavity, lends support to Turnheim's suggestion that the lesion is in fact produced by escape of cholesteatomatous matrix from the mastoid cavity it also renders unlikely the possibility that the cyst might arise as an implantation dermoid through a puncture of the intact skin over the mastoid process as a result of surgical trauma

There is a real value in the recognition of this disease, because nothing short of excision of sac, neck and source can be expected to rid the patient of a very troublesome complaint Knowledge of the fact that the neck of the sac does in fact run to the mastoid is a great help in the dissection In the first case reported, the author was looking for a neck to a branchial cyst, and in doing so must have unwittingly cut across the true neck leading upwards to the mastoid The result was failure, the cyst re formed within a short time The operation on the second case was correspondingly easier, and was rewarded with immediate success, because one knew where to search for the neck of the sac

The author has examined the world literature of the last 10 years, but can find no reference to the condition, except for Turnheim's one case report It is more than probable that similar cysts have been removed in the past, under a mistaken diagnosis of branchial cyst, or degenerating cervical lymph nodes.

Summary

Cholesteatoma cyst of the neck, first reported by Turnheim in 1955, results from the escape of cholesteatoma matrix from a mastoidectomy cavity or from an unopened mastoid air cell

Two further cases are recorded, and the suggestion is made that perhaps the condition is not so rare as perusal of otological journals would suggest

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CARCINOMA OF THE TRACHEA TREATED BY RADON SEED IMPLANTATION

By P. WINSTON* (London)

THE direct introduction of radon seeds to tumours of the trachea and bronchus is a procedure which is not often employed, although in inoperable or otherwise untreatable cases, considerable regression of the tumour with consequent symptomatic improvement may result.

The radon seeds consisting of gold tubing 5 millimetres long and 0·5 millimetres in diameter are introduced by means of the "gun" devised by F. J. Cleminson for the treatment of carcinoma of the œsophagus. The seeds are loaded with an amount of radon expressed as 1 or 2 millicuries. The intensity of the radiation is reduced to one half in about three and a half days and becomes clinically inert in two weeks. They may be implanted under direct vision through the appropriate endoscope under general or local anæsthesia. The seeds are distributed equally throughout the mass of tumour and in the type of case described a dose of 10 or 12 millicuries is employed. Their position is confirmed radiologically after insertion.

The following case of carcinoma of the trachea, illustrates the value of this procedure.

Mr. R. C. aet 69. Hardware warehouseman. First attended on *10.6.55* with a history of 3 months' cough, shortness of breath, and irritation of the throat. Previous history of having been investigated at another hospital in 1948 for anæmia when the hæmoglobin was found to be 38 per cent. and the anæmia was of the iron deficiency type. His weight was 10 st. 7 lb.

On examination there was obvious dyspnœa with inspiratory and expiratory stridor indicating tracheal obstruction. Indirect laryngoscopy showed the right cord to be immobile in the paramedian position and X-ray of the chest showed widening of the superior mediastinum. No abnormality was detected in the lungs. Tomograms of trachea and superior mediastinum confirmed the presence of a tumour in the right tracheal wall, with considerable narrowing of the lumen (Fig. 1).

On *28.6.55* a tracheoscopy was performed. An irregular area 1 cm. in length on the right wall of the trachea 21-23 cm. from the incisor teeth was found. A smooth nodule on the left wall of the trachea at the same level was removed for biopsy. Histological report was squamous cell carcinoma. The tumour was in the lower part of the trachea, inside the thorax and, therefore, surgically untreatable—the problem of reconstructing an excised trachea being as yet unsolved. The patient was, therefore, referred to the Royal Marsden

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* From the Institute of Laryngology and Otology.

Hospital for radiotherapy and received a course of deep X-ray therapy 5130 r between *1.7.55* to *25.8.55*.

On *11.10.55* he was seen in the radiotherapy clinic when the report was "Considerable improvement. Nil abnormal seen in larynx, both cords being now freely mobile. Breathing good and weight has increased. X-ray shows trachea relatively clear."

Further tracheoscopy was performed on *19.11.55*. This showed the swelling in the trachea to have increased in size and it was impossible to see beyond the swelling. Since he had already had a full course of radiotherapy it was decided to try the effect of implantation of radon seeds into the tumour. On *3.12.55* tracheoscopy was performed and 5 × 2 millicurie radon seeds inserted into the tracheal growth. Following this he was greatly improved, but on *4 6.56* he developed recurrence of dyspnœa and stridor. On *12 6.56* further tracheoscopy was performed and the trachea was found to be occluded by tumour except for a small lumen in the left anterior quadrant. Five more radon seeds (2 millicurie) were implanted into the tumour This led to considerable improvement. There was no dyspnœa or stridor and X-ray showed marked regression in the size of the tumour (Fig. 2).

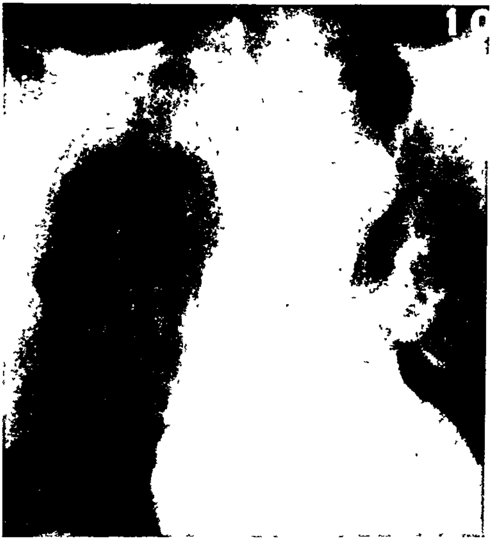

FIG 1

June 29th, 1955 Tumour obstructing trachea.

He kept well until *12.1.57* when he became dyspnœic and X-ray showed narrowing of the tracheal lumen. Tracheoscopy was performed and a further 5 × 2 m/c radon seeds were inserted into the tracheal growth. At the same time a swelling of the right tonsil was noted associated with an enlarged right tonsillar gland. Biopsy showed this to be a squamous cell carcinoma of the tonsil. A course of deep X-ray was given at the Royal Marsden Hospital with little effect. On *18.3.57* patient became very dyspnœic with inspiratory and expiratory stridor. There was extension of the ulceration of the right tonsil to the tongue and floor of mouth. On 27.3.57 tracheoscopy was again performed and showed there to be a very narrow tracheal lumen. Further radon seeds were inserted into the tracheal mass, and radioactive gold grains were inserted into the tumour originating in the right tonsil Shortly after this the patient developed bronchopneumonia and died on *19.4.57*.

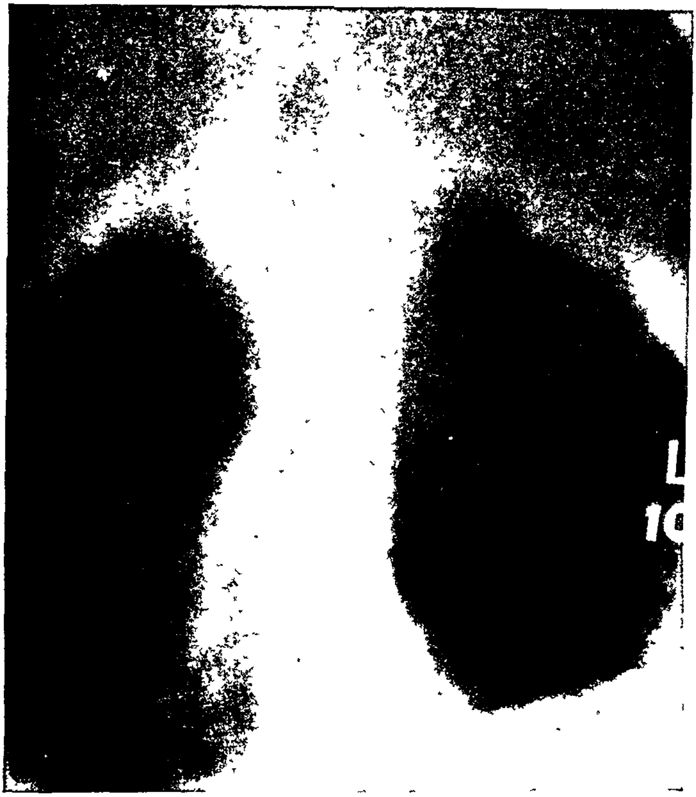

FIG. 2

August 20th, 1956 Result of treatment Radon seed still present

By means of repeated insertion of radon seeds an inoperable carcinoma of the trachea was kept under control and the patient remained at his work for 21 months after he first attended. At the end of this period he developed a second primary carcinoma in the right tonsil which considerably hastened his death.

Radioactive gold grains have now superseded the use of radon seeds in palliative treatment of carcinoma of the trachea and œsophagus because of the great accuracy with which they can be standardized and the dosage estimated. They are also uniform in dimension and their application is capable of much variation and development.

Summary

A case of carcinoma of trachea treated by radon seed implantation and kept under control for nearly 2 years. The development of a second primary carcinoma in the tonsil hastened his death.

Acknowledgments

This patient was under the care of Professor F. C. Ormerod to whom I am most grateful for all his help with this paper, to Dr. T. V. L. Crichlow and his staff for the X-rays and to Mr. D. J. Connolly for the photographs.

CLINICAL NOTES

THE USE OF PROLONGED INDWELLING POLYTHENE IN ANTRA IN AMBULANT PATIENTS

By J. FINE (Johannesburg)

It is common practice to introduce polythene tubing into infected antra for a limited period of up to one week, with the patient hospitalized.

In a recent series of 50 cases, polythene was left in for periods of up to 2½ months. The tube was cut short and tucked into the nasal vestibule out of sight, and the patient was sent back to normal duties in most cases.

Children and adults were dealt with in an identical manner and in very few cases was there intolerance or embarrassment. Polythene drainage was used thus in the following conditions:

1. Acute and chronic antral infections, including empyema, and antral infections complicated by
    - (*a*) Orbital invasion.
    - (*b*) Aural complications: middle-ear effusion with deafness, acute otitis media.
    - (*c*) Pulmonary complications—bronchitis, bronchiectasis, laryngitis.
    - (*d*) Postnasal drip.
    - (*e*) Other paranasal sinus infection: frontal, ethmoidal, sphenoidal.
2. Oro-antral fistula following dental extractions.
3. Sluder's and like facial neuralgias.
4. Advanced nasal stenosis from chronic allergic œdema of the inferior turbinates.

The following instillations were used for the appropriate condition after adequate preliminary nasal lavage.

1. Soluble antibiotics:
    Erythrocin Lactobionate.
    Tetracyclines.
    Penicillin.
2. For infective conditions hyaluronidase (1,000 units), in conjunction with antibiotics as indicated.
3. Alevaire was used when secretions were viscid especially in empyema and very chronic infections.
4. Lipiodol was used to further investigations in resistant cases or in cases suspect of tumour formation.
5. Procaine or Xylotox (lignocaine) in very tender antra requiring washout, and in facial neuralgias.

6. Various dyes—like triple dye, to test closure of oro-antral fistula and to test for effective ciliary function. The dye should be completely expelled within 24 hours in antra where cilia are still active.

7. Hydrocortisone acetate 5-10 mgm. in chronic allergic œdema resistant to all forms of therapy.

The following information can be derived from each and subsequent washouts.

(*a*) Healing progress by noting the nature of the secretions, e.g. their colour, quantity, odour, etc.

(*b*) Adequate and frequent specimens are available for investigations.

(*c*) Accurate prognosis can be determined. In recent cases the return fluid should be clear after 2 to 3 washouts. Persistent mucopus after one month suggests the existence of an irreversible state of the antral mucosa. Surgery is indicated in such cases. Lavage should be persisted with in such cases for a further period of 2 to 3 weeks, and if there is still no improvement, then radical surgery is the best course. Repeated antral washouts in this manner is even more effective than intranasal antrostomy, which operation will fail in those cases that have also failed to respond to polythene drainage. Patients subjected to radical surgery after such conservative treatment for a period of one month, seem to require less mucosal removal at operation and appear to respond most successfully to operation. This is probably due to the fact that they had a long period of pre-operative cleansing.

(*d*) Accurate localization of infection can be determined by noting the presence of mucopus in the nose and postnasal space immediately after antral washout. If such exists, then fluid displacement by the method of Proetz should be done. The presence of mucopus on the return flow will indicate infection in the ethmoidal and possibly sphenoidal cells. Treatment should then consist of combined lavage and displacement. Such combined treatment is frequently necessary in children.

Details of the Technique

(*a*) A proof puncture is done in the ordinary manner with a straight Lichtwitz cannula size 7·5 mm. The antrum is then flushed out.

(*b*) An 8 in. length of 1 mm. bore polythene tubing is threaded along the hollow of the cannula, which is then slowly withdrawn while continuous pressure on the polythene will cause it to suddenly bend and coil into the antrum. A length of 2-3 in should lie in the antral cavity. Incidently this procedure is an excellent test for novices doing proof punctures. It enables them to ascertain whether the cannula projects into the antral cavity before lavage. The tubing can be dipped into boiling water for half its distal length for softening before introduction, to facilitate curling up of the tubing. Test the patency of the tube in case of kinking. The projecting polythene is now cut short at the nasal tip and tucked into the vestibule, where it tends to stay put. Three days later, using a 10 c.c. syringe and No. 2 needle the cavity can be washed again. Lavage is not necessary more frequently than once daily or every two or three days. The return flow should be clear for a period of 7 days, during which period no lavage is employed, before the polythene is removed. Mild pressure neuralgia may develop in some cases after 1 or 2 weeks. In such cases rotate the tube to change its position or it may require removal. If desired, antibiotic instillations can be used, but lavage with plain saline has been found to be equally effective in most cases. Alevaire instillations are necessary with thick secretions.

Young children were found to be extremely co-operative and tolerant of this form of therapy. In most cases they were treated ambulant and older ones were sent back to school. By far the best results were found in younger patients.

Seven cases of facial neuralgias with no definite ætiological factor were treated. One cubic centimetre of hydrocortisone acetate alternating with lignocaine daily were instilled. Relief was prompt in three cases, and only of limited value in the rest. A larger series is necessary to evaluate this accurately.

It was interesting to discover how many cases still had mucopus after the first puncture, in acute and subacute infections.

So often one used to send these cases away. Could it be that they were incompletely treated?

The following are selected cases treated in the manner outlined above.

CASE I

A female child 8 years old. Thick mucoid middle-ear secretion on paracentesis on both sides. Polythene was introduced under general anæsthesia. Thick mucopus present in both antra. Drainage for 18 days. Instillation of 100,000 units of soluble penicillin. Result—full recovery of hearing. Antra cleared.

CASE II

Male, 12 years old. Orbital invasion from empyema antrum. High temperature, child ill. Polythene introduced under local anæsthesia, mucopurulent return. Systemic disinfection was provided. Hospitalized for 10 days. Polythene in for 21 days. Penicillin instillations daily. Full recovery.

CASE III

A male 3 years old with chronic œdema of inferior turbinates. Superadded purulent infection of antra. Polythene introduced for 14 days. Penicillin instillations alternate days. Hydrocortisone acetate instillation on last two days, with supportive anti-allergy therapy. Result—the infection cleared, and œdema of inferior turbinates was considerably reduced.

CASE IV

A male 36 years old with post traumatic acute frontal sinusitis, and œdema above his eye. Double proof puncture revealed 3+ mucopus both sides. Hospitalized for 10 days. Polythene drainage for 21 days. Instillations of erythromycin lactobionate alternate days. Systemic antibiotic disinfection. Good result after observation now for a period of 6 months.

CASE V

A male, 65 years old, with marked nasal stenosis from chronic nasal allergic rhinitis. Some superadded infection. One crystalline penicillin instillation (600,000 units). Marked dermatitis developed after two days. Penicillin vigorously washed out Anti-allergic and cortisone therapy instituted Nose better, but rash still severe Sent to bed and developed deep vein thrombosis Recovery of nose was good, but allergy to crystalline penicillin was responsible for a prolonged illness

CASE VI

Female, 32 Oro-antral fistula of 2 weeks duration, following extraction of upper right 4 Foul antral discharge on proof puncture Polythene drainage for a period of 3 months Only 12 washouts were necessary Result—complete closure of fistula

CASE VII

A male, 40 years, with oro antral fistula, 3 days duration Polythene in for 3 weeks Only one washout in first week, which was clear Further washouts may have expelled the clot in the opening Two washouts, one with triple dye to test closure of fistula, were done after 2½ weeks

CASE VIII

A male 40 years old with oro antral fistula of 3 months duration Foul mucopus present in his antrum with polyp and much granulation presenting in the mouth The infection cleared up rapidly, leaving a large clean epithelium lined fistula This case required a plastic repair and healed successfully Intra-nasal polythene only was used for drainage

CASE IX

Male, 50 years old, was a city lawyer with opaque left antrum discharging mucopus for many years He refused surgery from other otologists Polythene drainage for 2 months was necessary Hyalase, and alevaire instillations were used Result—surprisingly good No recurrence now after 1 year

CASE X

A city surgeon 50 years old He presented with an acute right empyema antrum No return flow with one cannula A second cannula was inserted and very thick viscid secretion mixed with thin mucopus flowed through first cannula Polythene drainage was necessary for a period of 14 days Instillation of hyalase, alevaire, and Erythromycin were used, in addition to systemic disinfection The patient rested for one day only

CASE XI

Headmaster, 52 years, with a very painful left empyema antrum following an attack of influenza He was hospitalized for 5 days Very thick mucopus present in the return flow Polythene retained for 18 days Xylotox instillations relieved painful pressure from lavaging Hyalase and penicillin were used Patient returned to school duties after 10 days with polythene, which was not noticed by any of the scholars

All three cases of oro-antral fistula were treated as ambulant cases, except in the one who had to have a minor plastic repair at a later date. They had all been seen by plastic and maxillo-facial surgeons who contemplated surgery sooner or later, with the usual intranasal antrostomy. All these cases were given supportive therapy for one week.

It is apparent that the management of cases in this manner amounts to a test of the reversibility of antral and adjacent mucosa, back to normal. In cases of long standing where healing occurs, and the mucosa presumably reverts to a healthy state, a prolonged period of observation is necessary, as recurrences are frequent. These cases are likely to flare up again and remain infected after coryza, influenza or debilitation from any cause. This vulnerability will persist for some time in numbers of cases, while there is no evidence of activity and a clear return on proof puncture. A course of drainage may have to be repeated to stabilize the condition.

Not a single case in this series appeared to have been aggravated or complicated from this treatment.

There is no need to state that supportive measures were advised when indicated, particularly in the acute infections.

Swimming, and more especially diving and jumping into filtered or sea water was prohibited during treatment. Caution in this respect was advised in all cases for a period after treatment had ceased.

It is evident that patients who have had antrostomies done cannot be dealt with in this manner, unless the opening had closed up again.

As regards cases of oro-antral fistula it appears that this form of treatment holds out good hope for a more conservative line. This is particularly so for the more recent cases. Even chronic cases benefit considerably as the field can be given a prolonged course of pre-operative cleansing, thus giving hope for lesser incidence of failure of surgical repair. In all cases the polythene should be kept in for a prolonged period, and it is surprising to find that a severely infected antrum with foul secretions will respond to even a single washout. It is also necessary to X-ray cases to eliminate the presence of roots or sequestra. One cannot help feeling that every case of oro-antral fistula should be referred to a rhinologist first for such therapy before any form of surgery is commenced.

USE OF STREPTOKINASE STREPTODORNASE IN LARYNGO-TRACHEOBRONCHITIS

By A RUTANA (Liverpool)

It is well-known that tracheotomized children with non bacterial laryngotracheobronchitis are very difficult to manage This is due to crusting of the trachea and bronchi which always occurs in this particular kind of infection Most likely the crusting of the air passages is due to changes in the mucus secreting glands in the air passages This crusting is not due to tracheotomy as it occurs previous to operation Bronchoscopy, performed for removal of the crusts, tends to denude the mucosa of the trachea and bronchi and thus increases the crust formation

This is a case of acute laryngo tracheobronchitis treated with Streptokinase-Streptodornase, prepared by Wellcome Research Laboratories A girl, aged 2 years, was admitted to hospital on *September 26th, 1957*, She was perfectly well until the day before admission when she developed a "croupy" cough and nasal discharge At the beginning the cough was not troublesome, she was playing the whole day However, about 2 a m she woke up and her breathing was very rapid and she was very hot She was given a drink, which she vomited, and the mother went for the doctor who admitted her to hospital

On admission she had a temperature of 102°, was extremely ill, very cyanosed with a shallow irregular breathing and marked rib and sternal recession She was very restless There was no history of a foreign body having been inhaled There was diminished air entry throughout, due to obstruction of airway, but no adventitious sounds apart from mucus in the trachea She was treated with Oxygen and *Alevaire Tracheotomy* was advised *On direct laryngoscopy*, a supraglottic œdema was found to be present

Tracheotomy was performed, and about 2 c c of yellow thick pus was aspirated from the trachea This was sent for examination and direct film showed debris only, no organisms and culture showed no bacterial growth obtained She was then put back into the Oxygen tent with Alevaire and suction of the tracheotomy tube was advised She was put on Penicillin and Streptomycin injection

Her general condition was quite satisfactory, her breathing was good and her chest clear up to *September 29th, 1957* She then became rather distressed and dyspnœic As the tracheotomy tube was clear she was taken for bronchoscopy This was performed on *September 30th, 1957* Bronchoscopy showed blood stained secretion, mucus with pus and thin crusts in the trachea causing obstruction

The same evening she became obstructed again due to crust and secretion This necessitated another bronchoscopy, on which a considerable amount of blood stained crusts were removed from the neighbourhood of the carina

Her condition then remained good until the following afternoon when she developed sternal recession and started gasping for breath. There was a diminished air entry at the right base and chest X-ray showed partial collapse of the right middle lobe, with displacement of the heart to the left. She had another bronchoscopy done and a considerable amount of dry crust was removed from both bronchi and trachea.

After this bronchoscopy the child was not as well as after the same procedure before. There was a very poor air entry on both sides. She was put back again in the Oxygen tent with Alevaire.

The following morning her general condition had deteriorated considerably. She was very restless and excited. There was a marked recession of the ribs. The tracheotomy tube was absolutely clear.

Her condition was then considered very grave. Her pulse was rather poor and rapid. A solution of Streptokinase and Streptodornase was made, 3 c.c., and injected through the tracheotomy tube into the trachea and sucked out in about 10 minutes. Dry crusts and mucus were removed with the sucker followed by a yellow pus. This was followed in about fifteen minutes by another injection of 3 c.c. of a solution of Streptokinase and Streptodornase, following which more dry crusts and pus was sucked out. The air entry improved immediately and the general condition of the child was completely changed for the better.

She then fell asleep and remained all right for about five hours. She became obstructed again and another 3 c.c. of Streptokinase and Streptodornase were injected. Mainly mucopus was aspirated.

Her subsequent post-operative course was satisfactory and she was discharged from hospital on *October 13th, 1957*.

This article is written to draw attention to the efficiency of this type of treatment.

Acknowledgments

I wish to thank Miss A. E. McCandless, Pædiatrician, and Mr. John McFarland, E. N. T. Surgeon, for the permission to publish this case.

GENERAL NOTES

THE Editors are anxious to improve the clinical aspect of the Journal and would welcome more reports of interesting cases These clinical records need not be at all elaborate and it would be unnecessary to go minutely into the ætiology or the literature of the subject Senior Registrars have unique opportunities for seeing a large variety of cases, many with unusual features It is in many ways regrettable that these should go unrecorded It is hoped that all those in charge of clinics will help us in this matter and see that such cases are not overlooked

OTOLARYNGOLOGY LECTURES 1957-58

THE following lecture, arranged jointly by the Royal College of Surgeons of England and the Institute of Laryngology and Otology, will be delivered in the Lecture Hall of the College, at 5 30 p m

1958
July 3rd Mr Norman C Tanner Haemorrhage from the Œsophagus

INTERNATIONAL CONGRESS ON THE MODERN EDUCATIONAL TREATMENT OF DEAFNESS

AN International Congress on the Modern Educational Treatment of Deafness will be held at the University of Manchester from July 15th 23rd, 1958

The Congress is being organized with the approval and support of the Council of the University, with financial help from the Ministry of Education, the National College of Teachers of the Deaf, and the Rathbone Trustees, and with the co-operation of the British Council and the Alexander Graham Bell Association for the Deaf, Inc , Washington, D C , U S A

The Congress is being planned to bring together educators, otolaryngologists, pædiatricians medical officers, audiologists and educational psychologists, to consider and discuss the significance, to the future development of their work of the results of modern research on problems of deafness in childhood

Arrangements are being made to offer accommodation to members of the Congress in University Halls of Residence The cost of this type of accommodation is expected to be about £1 2s per day, including bed, breakfast and dinner The University Refectory will be available for meals during each day

Forms of application for membership of the Congress will be supplied upon request, to Professor A W G Ewing, Department of Education of the Deaf, the University, Manchester 13, England

General Notes

FACULTÉ DE MÉDECINE DE PARIS

Chaire de Clinique Oto-Rhino-Laryngologique. Professeur A. Aubin. Cours de Technique Chirurgicale—La Chirurgie de la Surdité. Lundi, 9 au Samedi, 14 Juin, 1958.

Apply Pavillon Isambert, Hôpital Lariboisière, 2 Rue Ambroise-Paré, Paris.

ROYAL SOCIETY OF MEDICINE

SECTIONS OF LARYNGOLOGY AND OTOLOGY

Summer Meeting in Edinburgh, June 27th, 28th, 1958.

Friday, June 27th.

9.30 a.m. Scientific Meeting: Royal College of Surgeons of Edinburgh, Nicholson Street. *Discussion:* "Acoustic Neuroma". Professor Norman Dott. Dr. C. S. Hallpike. Dr. D. Dix.

1 p.m. Luncheon for Members and Guests. Carlton Hotel, North Bridge. Price 15s. inclusive of wines and gratuities.

Afternoon. Garden Party by invitation of the Presidents.

7.15 for 7.45 p.m. Dinner in The Royal College of Surgeons of Edinburgh. Tickets Two guineas each inclusive of wines.

Saturday, June 28th.

9.30 a.m. Scientific Meeting: Royal College of Surgeons of Edinburgh. *Discussion:* "The dysphonias and the scope of speech therapy and their treatment". (Illustrated by recordings). Mr. Maxwell P. Ellis. Mr. L. Willmore.

12.30 p.m. Meeting ends.

For details of accommodation write to the Secretaries of the Sections.

Registration Fee of 10s. will be made for each Member to cover overhead expenses.

Members wishing to attend the Meeting should apply *as soon as possible.* Bookings at hotels must be made through the Royal Society of Medicine and *not* directly with the hotels. Accommodation is limited and early application should be made.

Applications should be accompanied by a cheque *made payable to Dr. J. F. Birrell* to cover the registration fee, lunch and Dinner tickets, but *not* for hotel accommodation.

N. L. CRABTREE, Honorary Secretary, Section of Laryngology.
LIONEL TAYLOR, Honorary Secretary, Section of Otology.

The Journal of
Laryngology and Otology

(*Founded in* 1887 *by* MORELL MACKENZIE *and* NORRIS WOLFENDEN)

July 1958

THE ÆTIOLOGY, DIAGNOSIS AND SURGICAL TREATMENT OF PHARYNGEAL DIVERTICULA

By F. BOYES KORKIS (London)

INTEREST in the surgical treatment of pharyngeal diverticula has been re-awakened by Dohlman (1949) who advocates endoscopic, diathermic division of the ridge separating the œsophagus from the diverticulum. This technique which preserves the pouch, offers the obvious advantages of simplicity and speed. Whether total pharyngo-diverticulectomy should be discarded in its favour is debatable as the results of radical treatment are gratifying. Having had no first-hand knowledge of the endoscopic method, and only a limited experience of diverticulectomy, the author will resort to the published results of other surgeons in advocating external operation.

Historical Summary

Pharyngeal diverticula have interested several branches of our profession since Ludlow, of Bristol, in the year 1769 gave the first accurate pathological account of the condition. He found an abnormal dilatation of the posterior pharyngeal wall when he performed a post-mortem on a patient who had suffered from dysphagia during life. A century later, in 1864, Kunge removed a diverticulum but the patient died. Hamburger, in 1871, and Niehans, in 1884, were also unsuccessful. Zenker, in 1876, as a result of a detailed study of the condition, recommended the removal of the sac. It remained for Wheeler, of Dublin, to report the first successful excision in 1886, the operation being performed in 1885.

In those days total excision of the diverticulum carried a high operative mortality due to sepsis. Because of this danger the operation fell into disrepute. However, Goldmann, in 1909, proposed a two-stage procedure as a safeguard against infection. Some eminent surgeons (Lahey, etc.) still advocate this method in our antibiotic era, but nowadays most have reverted to the one-stage operation.

External operation with removal of the sac in one or two stages had thus become the accepted form of treatment for large diverticula producing symptoms, as other operations designed to invert the fundus of the pouch (Girard, 1896; Halstead, 1904), or to displace it upwards (Hill, 1926) had been given up owing to the frequency of recurrence.

Incidence

The condition is uncommon and British surgeons have not been able to gain as large an experience as some in other countries. In the United States of America it would appear to be more frequent, to judge by the larger number of cases collected by individuals. Shallow and Clerf (1948) have estimated the incidence at the Jefferson Hospital at 1 in 800 operations, or at 1 in 1,400 admissions. Comparative statistics do not reflect the true incidence as surgeons who possess extensive experience of the condition will naturally attract more cases. This tendency is more pronounced on the other side of the Atlantic where there is a greater trend towards ultra-specialization. The general incidence in Britain must be much lower. Before the establishment of a special department at Hillingdon Hospital in the 10 years from 1942 to 1951, there were 119,523 admissions, yet only one of these was diagnosed on discharge as a pharyngeal diverticulum. Now one is seeing perhaps 2-3 cases per year. Comparative figures of the incidence must, therefore, be looked upon with reserve.

Personal Material

A total of 24 have been seen in the past 11 years, but owing to failure to trace 5 patients, adequate information is available for only 19. This limited personal experience relates to 15 males and 4 females whose ages ranged from 34 to 78 years, the average age at the time of examination being 59 years. Thirteen of the nineteen were treated by diverticulectomy; one by repeated œsophageal dilatation; and 5 required no specific treatment other than general advice on dietary habits.

Anatomical Types

Pharyngeal diverticula arise in 3 regions, viz., posteriorly, laterally and postero-laterally. The anterior type which Hurst (1925) described and called "antero-pharyngo-œsophageal" does not occur. Hurst himself retracted his former opinion by admitting his error in mistaking barium in the normal vallecula for a diverticulum (Morley, 1945). A diverticulum arising from the vallecula has been reported by Kaufman (1956), but this was a lateral diverticulum.

(1) *Posterior.* This is the most frequent type. It arises as a herniation of the pharyngeal mucosa through the fibres of the inferior constrictor muscle. The arrangement of the musculature in this region accounts for the site of election. The inferior constrictor consists of two distinct portions, an upper oblique, and a lower horizontal part. The horizontal fibres run from side to side to form a muscular sling which on contraction acts as a sphincter to shut off the pharynx from the œsophagus. This portion of the muscle is an entity distinct from the oblique upper portion, and the name cricopharyngeus muscle is a good descriptive term for it. The diverging muscles leave a relatively weak, unsupported area posteriorly through which the pharyngeal mucosa herniates. Thus, a true posterior pharyngeal diverticulum arises above the crico-pharyngeus muscle. They are usually considered to be acquired. All 19 cases in this series were of this type.

(2) *Lateral.* Lateral pouches according to Atkinson (1952) are of three types:

- (*a*) Pharyngocœles produced by prolonged excessive pressure on a weak area in the lateral pharyngeal wall, i.e. due to increased air pressure in, for example, glassblowers.
- (*b*) Mechanically produced pouches acquired artificially by pressure on the lateral pharyngeal wall of the hypopharynx by foreign bodies, such as coins, digital pressure, etc., i.e. coin swallowers.
- (*c*) Congenital, representing a persistence for the second, third, or fourth branchial pouches. For example, in Kaufman's case a small lateral pouch arose from the vallecula, and endoscopy revealed a small slit-like groove in the right vallecular area.

(3) *Postero-lateral* pouches have been described as occurring below the cricopharyngeus between this structure and the uppermost part of the œsophagus. Moynihan (1927) found two such cases reported in the literature and 2 cases out of the 186 reported by Shallow (1948) were thought to have originated through a postero-lateral slit.

Aetiology

The concensus of present-day opinion favours an acquired origin for the common posterior variety. In many cases no cause can be found clinically and the explanation remains hypothetical. The outpouching is usually considered to arise as a result of pressure from within, the diverticulum being of pulsion type. Neuro-muscular inco-ordination caused by delayed relaxation of the crico-pharyngeus muscle during the second stage of swallowing is held to be responsible by many. Sir Victor Negus (1950) believes that tonic contraction of this muscle prevents the downward passage of the bolus. The relatively weak posterior pharyngeal wall gives way owing to the long continued intrapharyngeal pressure and eventually herniation results. Amongst many others, the late Frank H. Lahey subscribed to this view.

Although the theory of neuro-muscular inco-ordination is widely accepted, it does not explain the following clinical facts:

(1) Following operation the condition is cured while the alleged cause remains. (Dunhill, 1950.)

(2) A familial history has been noted occasionally. Diverticula occurring in several members of one family have been reported by Björk (1952), Dunhill (1950) and others.

(3) An association with multiple diverticula in other anatomical regions, as recorded by Solis-Cohen *et al.* (1956), and by Björk (1952).

(4) An association with multiple herniae in other regions, as in a patient reported in this series who had previously been operated upon for multiple abdominal herniae. Solis-Cohen's patient had a hiatal hernia, as well as multiple diverticula.

(5) The absence of symptoms and disability in some patients who have demonstrable pharyngeal diverticula. (Dunhill, 1950.)

(6) Absence of the development of a diverticulum in most cases of long-standing stenosis.

(7) Dysphagia in middle-aged women is more common that in middle-aged men, but pharyngeal diverticula occur about 4 times as often in males as in females. This reversal of the incidence suggests that other factors besides neuro-muscular inco-ordination must be operative.

The question is what are these factors? Organic disease producing stenosis is a well-known cause. Wolfsohn reported such a case before the clinical section of the Royal Society of Medicine in 1950. One of the patients in the present series dates her symptoms from swallowing a chicken bone; demonstrable stenosis was followed by the development of a pouch. Likewise interference with the nerve supply to the hypopharyngeal muscles from any cause might lead to a weakening of muscular tone. Three of my patients had well marked radiological evidence of pulmonary fibrosis, three had bulbar palsy and one suffered from a complete left recurrent nerve paralysis pre-operatively. Extensive investigation failed to reveal the ætiology of this nerve lesion. As will be seen from Table I, a possible clinical cause of dysfunction in the crico-pharyngeal region was found in 11 of the 19 patients In some the association is a matter of conjecture but in others the connection between the two events seems fairly certain

TABLE I

POSSIBLE ÆTIOLOGICAL FACTORS IN 19 PERSONAL CASES

Associated Pre Op Condition	Number	Remarks
Pulmonary Fibrosis	3	1 T B + ve ? *Primary* ? Secondary to Spill
Bulbar Palsy	3	No operation
Left Recurrent Laryngeal Paralysis (Cause unknown)	1	Present pre operatively
Quinsy and probably Parapharyngeal Abscess with Fibrosis	1	Dysphagia dates from Quinsy 2 years previously
F B Perforation of Cervical Œsophagus with subsequent Stenosis	1	Definite relation drip Pouch developed in 3 months
Multiple Herniae	1	? Congenital muscular weakness
Cervical Disc Lesion C 6 C 7	1	? Related
Unknown	8	? Neuro muscular inco ordination
Total	19	

It is distinctly uncommon for a pouch to develop in association with stenosis, or a nerve lesion Some special predisposing factor must be postulated to explain why the same exciting factor leads to herniation in some patients and not in others (Perzik, 1953)

It seems likely that there is a congenital posterior defect between the fibres of the inferior constrictor muscle, the area remaining as a potential site for herniation (Perzik, 1953) This only takes place in the presence of an exciting factor which produces obstruction, this may be neuro-muscular dysfunction or organic disease A predisposing congenital weakness is in my opinion the most likely theory which fits in with the clinical facts

It is often argued that a congenital cause can be dismissed as pharyngeal diverticula are not found in young children This is no longer a valid argument for Brintnall and Kridelbaugh (1950) have described 2 cases of posterior diverticula found at operation in babies aged 8 and 2 days respectively These large posterior, midline diverticula must have been present as a congenital deformity from birth Neuro-muscular inco-ordination has also been reported in a newly-born baby by Morgan (1956) No abnormality was found at post-mortem Birth trauma was postulated as the cause, the forceps delivery disturbing the fibres of the recurrent laryngeal nerve If this theory is correct, dysphagia, produced by neuro-muscular inco-ordination in early life, could initiate a symptomless pulsion diverticulum Progression would then cause the development of typical symptoms later

Aetiological Classification

By accepting the theory of combined congenital predisposing and acquired exciting causes many debatable points are resolved. It is useful to subdivide pharyngeal diverticula accordingly:

(1) *Primary Congenital*
  (*a*) Lateral. Persistence of embryological remnants (very rare).
  (*b*) Posterior I. Manifest. Symptoms simulating œsophageal atresia arise in infancy.
    II. Latent, or Occult. Small symptomless diverticula in infancy. Progress with symptoms in later life. (? Congenital neuro-muscular dysfunction.)
(2) *Secondary Acquired*
  Primary congenital predisposition.
  Posterior: The common diverticula which develop only when an exciting factor (neuro-muscular inco-ordination, nerve lesion, stenosis, etc.) is operative.
  Postero-lateral: Herniation below the cricopharyngeus (? Congenital).
(3) *Primary Acquired*
  Lateral: Increased intra-pharyngeal pressure, or trauma, i.e. traumatic pharyngocœles.

Clinical Classification

The system of clinical staging suggested by Lahey and Warren (1954) has been applied to the cases in this series. It serves the useful purpose of pointing the way to the correct treatment for a given patient. Three stages are recognizable as distension and the weight of the contents of the pouch increase its size.

Stage 1

A bulge of the mucosa and submucosa through the pharyngeal dimple but no sac. The sensation produced is that of a foreign body which leads to "hawking" in an attempt to dislodge it.

Stage 2

A globular sac develops but there is no rotation of the upper œsophagus. Retention of food and mucus occurs but there is no obstruction.

Stage 3

Rotation of the œsophageal introitus causes the mouth of the diverticulum to be dependent so that the hypopharynx runs directly downwards into the pouch. Obstructive symptoms are now present.

The distribution of the present series of 19 patients is shown in Table II.

Clinical Features

The symptoms and signs are detailed in Tables III and IV. The usual symptoms of regurgitation and dysphagia are well known. The importance

TABLE II

Stage	Description	Symptoms	No of patients
1	Simple Bulge	Foreign body and hawking	4
2	Globular sac	Retention	2
3	Elongation of sac with rotation of the œsophagus	Obstruction	13
		Total	19

TABLE III

SYMPTOMS IN 19 CASES

Stage 3 (13 cases)	
Dysphagia (solids more often than liquids)	13
Regurgitation	13
Cough	8
Gurgling noises	5
Hoarseness	3
Lump in Neck	3
Loss of Weight	3
Stages 2 and 1 (6 cases)	
Dysphagia (3 cases Bulbar Paralysis)	4
F B (foreign body) (sensation of)	3

TABLE IV

SIGNS IN 19 CASES

Stage 3 (13 cases)	
Emaciation	2
External swelling	2
Gurgle on pressure	6
Retention Frothy Fluid in Hypopharynx	9
Congestion Vocal Cords (Laryngitis)	2
Left Vocal Cord Paralysis	1
Barium Swallow Single Sac	12
Bilobed Pouch	1
Stage 1 and 2 (6 cases)	
Paralysis 9th 10th 11th or 12th Cranial Nerves with Retention	3 (Bulbar Palsy)
No clinical signs	3

TREATMENT ADOPTED (19 Cases)

Observation only		2 (Stage 1)
Dietary measures including tube feeding		3 (Stage 1 and 2)
Endoscopic dilatation		1 (Stage 2)
External operation	Total	13 (Stage 3)
Two Stage		2
One Stage		11

of cough, the third symptom in order of frequency, has been insufficiently stressed In 2 cases cough was the leading symptom and in another it preceded the dysphagia by several years Cough arises in 3 ways

(1) As a protective reflex to prevent the retained contents from spilling

over into the respiratory tree. In this instance cough is related to eating or to drinking, especially when the patient swallows in a hurry and is often worse at night time due to the recumbent position. In one patient the cough was so violent that it produced a choking sensation.

(2) By aspiration. Recurrent pneumonitis leads to bouts of coughing with freedom between the attacks. A careful clinical and radiological chest examination is called for to exclude pneumonitis or fibrosis. The radiological changes are usually basal, but in 3 patients apical fibrosis was also present. Recurrent broncho-pneumonia may necessitate emergency operation, as in the following case:

A female patient aged 72 was admitted with a diagnosis of recurrent "bronchitis". Antibiotic therapy failed to control the attacks and she became gravely ill. Radiological examination revealed a large diverticulum which was removed in two stages on account of her poor general condition. She was restored to health and did not suffer any further bouts of so-called "bronchitis".

(3) By pressure on the recurrent laryngeal nerve. In the case reported by Dunhill the cough was incessant. The question arises, can pre-operative recurrent laryngeal nerve paralysis be produced by this mechanism? Investigation of the patient in this series revealed no other cause. As this particular patient suffered from a persistent cough which preceded his hoarseness, this is possible.

It is unusual to obtain a history of recurrent swelling on the left side of the neck after food, but this was the main complaint of one male patient. Examination revealed a prominent bulge in his left supraclavicular region. Gurgling noises on pressure may sometimes be elicited after swallowing a glass of water, whereas they are absent prior to this. The most common clinical finding is that of retention of saliva in the hypopharynx.

A barium swallow examination is necessary to demonstrate the diverticulum. This may show a single sac or a bilobed diverticulum. Bilobed diverticula are rare (Jesberg, 1954), but one male patient, aged 71, had this type of pouch. The bilobed nature was not evident in antero-posterior X-rays but a notch was noted in the lateral film. At operation the bilobed character was greater than was expected from the X-ray appearances, the cleft extending well towards the pharyngeal wall.

Although there have been reports of carcinomata occurring within pharyngeal diverticula, this complication was not encountered in the present series. Lahey and Warren (1954) point out that irregularity of the outline of the barium-filled pouch is suspicious. Endoscopic examination is, therefore, always advisable.

An association with peptic ulceration has been noted by Slade (1954) who has reported 3 cases. No such association was found in this series.

Treatment

When a diverticulum is small, and is producing few symptoms conservative treatment is indicated. When it is secondary to advanced, organic neurological disease, such as bulbar palsy, operation is not advisable. Of the 19 cases, 6 were treated conservatively and 13 patients with Stage 3 diverticula were subjected to external operation (Table IV).

Amongst measures giving considerable relief in minor herniations are lavage with alkaline solutions after meals, careful attention to the diet and feeding habits, and periodical endoscopic dilatation of the œsophageal introitus (crico-pharyngeal sphincter). In early stage one cases regular observation is the only measure necessary. This was done in 2 instances. With bulbar paralysis (3 patients) it was felt that regular tube-feeding offered the best means of maintaining nutrition, as the herniation played only a small secondary part in the production of dysphagia.

The female patient who was treated by dilatation was kept swallowing comfortably for 5 years until she died of unrelated disease (carcinoma of the bladder) this year. Her diverticulum was border-line, Stage 2-3. Dilatation was required twice yearly and was repeated as soon as she had a return of symptoms.

Operative excision was performed on 13 patients, the two-stage operation being done on account of their poor general condition. Today these would probably have been treated by diathermic division of the septum. Poor risk patients are the chief indication for this type of surgery in my opinion.

In the past there has been a tendency for some physicians to treat large diverticula conservatively on account of the supposed risks of the operation. The actual incidence of fatal mediastinitis is in fact extremely low when a meticulous one-stage or two-stage operation is performed. An

TABLE V

ONE-STAGE OPERATIONS

Authors	Year	Total No of operations	Deaths	Fatal mediastinitis
Shallow and Clerf	1948	186	5 (3%)	0
Sweet	1956	67	1	0
Harrington	1945	115	0	0
Jackson and Norris	1955	53	1	0
Mortensen *et al*	1955	339	3 (0.9%)	0
Morley	1945	21	0	0
Dunhill	1950	19	1	0
		799	11 (1.4%)	0

KORKIS, F. B.—*Recent Advances in Oto-laryngology*, 3rd edition analysis of 799 one-stage operations, reported by 7 different surgeons, showed an overall death rate of 1·4 per cent. (Table V.) No deaths were due to mediastinitis. In 393 two-stage operations (done by 3 surgeons) the operative mortality was 0·76 per cent., one death being caused by mediastinitis (Table VI).

TABLE VI.
TWO-STAGE OPERATIONS.

Authors	Year	Total No. of operations	Deaths	Fatal mediastinitis
Harrington	1945	25	1	0
Lahey and Warren	1954	365	2	1
Dunhill	1950	3	0	0
		393	3 (0·76%)	1

KORKIS, F. B.—*Recent Advances in Oto-laryngology*, 3rd edition.

Dohlman's method has found favour in some quarters on account of the supposed dangers of external operation, but these figures refute this idea. Moreover, local complications, such as post-operative stenosis, recurrence and damage to the recurrent laryngeal nerve are rarely encountered by experienced surgeons, as shown by Harrington's (1945) series of 140 operations (Table VII). There is, therefore, no need to abandon a well established operation giving a high percentage of permanent trouble-free cures.

TABLE VII.
POST-OPERATIVE COMPLICATIONS (Harrington).

Complication	One-Stage Operations (115)	Two-Stage Operations (25)
Recurrence	2	3
Temporary fistula	5	6
Temporary hoarseness	3	3
Permanent paralysis of recurrent laryngeal nerve	1	1
Stenosis requiring dilatation	5	5

KORKIS, F. B.—*Recent Advances in Oto-laryngology*, 3rd edition.

Personal Technique

When total diverticulectomy has been successfully performed, it matters little in the long run whether the one stage or the two-stage method is used as both are satisfactory and safe. The choice is a personal one My own preference favours the one stage technique This makes for rapid post-operative recovery with minimum discomfort to the patient

Careful pre-operative preparation prevents the retention of food in the pouch The patient carries out lavage after each meal for one week prior to admission by rapidly gulping water and regurgitating it Three days before operation he is admitted during which time further lavage is performed by the nursing staff who use a small Ryle's feeding tube On the morning of operation this is passed *viâ* the nostril As a large amount of saliva can collect in the pouch in a short time, the tube is retained in order to aspirate the contents immediately before operation in the anæsthetic room

Antibiotic cover is used routinely Systemic penicillin is started 24 hours pre-operatively Deep breathing exercises are also given by a physiotherapist in the interval between admission and operation

General anæsthesia is employed The first step is an endoscopic examination using Negus's œsophageal speculum Should a suspicious area of ulceration suggest the possibility of carcinoma, the external operation would be deferred until the result of biopsy was available, but this has not been necessary to date The Ryle's tube is replaced by a large Jacque's feeding tube which is guided into the œsophagus and the diverticulum is lightly packed with ribbon gauze This technique dispenses with the need for further endoscopic guidance during the course of the diverticulectomy and is simple

In 3 early cases the classical incision along the anterior border of the sternomastoid was used but this produced permanent visible scarring A left transverse collar incision has now replaced it and has been found to be highly satisfactory in 10 cases This method of approach gives adequate access and leaves an almost invisible scar Good retraction with ligation and division of the middle thyroid vein allows a free exposure of the packed diverticulum It is rarely necessary to divide the omohyoid muscle and the inferior thyroid artery The pouch is gently teased out of its bed, freed up to its neck and delivered into the wound A large vein on its posterior aspect often requires ligation to prevent troublesome oozing

It is necessary to demonstrate clearly the junction of the diverticulum with the pharynx The feeding tube which can be felt through the wall is a useful guide The neck of the pouch is not clamped for this produces a crushed, devitalized mucosal edge which is liable to break down later For a similar reason a purse string suture around the neck is avoided The neck of the sac is cleanly divided level with the wall of the œsophagus

An eyeless needle carrying the finest intestinal catgut is used to close the pharyngeal opening A continuous inverting suture is employed but the thinness of the mucosa makes it imperative to handle it the instruments and the catgut with the greatest delicacy Water-tight closure is the aim Any muscle fibres which are often so thin as to be almost nonexistent are then approximated with interrupted sutures. The sternomastoid muscle is tacked down to the prevertebral area to shut off the carotid sheath and finally the wound is closed by a 2 layer approximation of the platysma and of the skin. Drainage is avoided. When the wounds were drained, post-operative fistulae were more likely. Four out of six patients who were thus treated subsequently developed leakage. Since changing the technique this complication has been largely overcome.

Results

Pharyngeal diverticulectomy requires considerable experience and it is natural that in the commencement more difficulties were encountered. In assessing the value of external operation, the results should be judged by the complications attending operations performed by surgeons thoroughly experienced in this work. The incidence of complications in this small personal series in no way reflects the excellent results which are being achieved by more experienced operators. Rather it is a measure of one's own progress as most of the complications (Table VIII) were encountered in the earlier operations. Of the last 7 cases, 6 had a smooth uninterrupted post-operative course. The wounds healed by primary intention and the patients were fit for discharge from the hospital on the 14th to the 16th day. The operating time now averages only about 45 minutes.

TABLE VIII.
RESULTS.

Post-Op. Complication	One-Stage (11 cases)	Two-Stage (2 cases)
Fistula	2 (1 T.B.)	2
Lt. recurrent laryngeal nerve paralysis (Late—not immediate)	1	0
Local infection	2	2
Minor stenosis	1	1
Recurrence	0	0

One local complication was often accompanied by another in the same patient. Thus, local infection and fistula formation frequently co-existed. Both patients who had a two-stage resection developed fistulae, whereas only 2 out of 11 one-stage excisions did so.

As a search of the literature has failed to find a similar case, the opportunity is taken to record the development of a tuberculous fistula post-operatively. A male, aged 46, had a two year history of cough with "white" sputum, and a 3 month history of regurgitation of undigested food. Barium swallow showed a large posterior diverticulum and the chest X-ray revealed patchy consolidation in both lungs, including the apices.

Serial bacteriological examinations of the sputum failed to demonstrate tubercule bacilli and in consequence the lung changes were thought to be caused by fibrosis following upon aspiration. A one-stage diverticulectomy was performed without difficulty but on the 11th post-operative day the wound broke down and a fistula developed. After repeated packing the pharyngeal opening closed but a discharging sinus persisted. Granulation tissue removed from this sinus was tuberculous in type and tubercle bacilli were seen in it and later found in the sputum. After a long course of streptomycin and repeated packing of the sinus, complete healing was finally attained some 18 months later. Since this time this patient has remained well for the past 4 years.

Local infection caused by pyogenic organisms often resolves with conservative therapy, but when a local abscess develops early incision may obviate secondary fistula formation. Fistulae normally close spontaneously if the tract is lightly packed but if they persist over 2 months they are closed by a secondary operation. In such cases it is important to exclude stenosis below the internal opening of the fistula. Dilatation would then be necessary to overcome the obstruction.

Conclusions

In the light of past experience, one-stage resection will remain the operative procedure adopted for reasonably fit patients who require operation. Total excision of the diverticulum may be exacting, but it is safe and satisfactory, giving excellent long-term results in experienced hands. It is not an operation for the occasional cervical surgeon as familiarity in handling soft tissues is essential. Gentleness in dealing with the delicate mucosa and extreme care in suturing are the handmaidens of success.

Endoscopic division of the septum may prove to be the method of choice for the desperately ill patient but retention of the sac carries definite risks. Infection can develop in the diathermized tissues and carcinoma may occur in the future, as malignancy has been reported following successful diverticulopexy. In my view, the surgeon has not the same measure of control as in an exact external operation. For these reasons this new method of treatment should not supersede pharyngo-diverticulectomy for routine stage 3 diverticula.

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THE ÆTIOLOGY, DIAGNOSIS AND SURGICAL TREATMENT OF PHARYNGEAL DIVERTICULA

By M. SPENCER HARRISON (Lincoln)

IN the early stages of muscular inco-ordination the cricopharyngeus gives way and the patient usually complains only of some hold up in the laryngeal area during swallowing, but the amount of discomfort and pain are not always in proportion to the size of the diverticulum and much discomfort may occur even in the very early stages, at times even before the actual formation of a diverticulum where there is merely a small depression above the mouth of the œsophagus accompanied by a spasm of the cricopharyngeus muscle. It is interesting that what are probably the earliest symptoms of the condition are by no means typical of the established diverticulum. The patient, during times of mental stress and pressure of work, finds that he has difficulty in passing food through the upper œsophagus and has a feeling of tightening in the throat, usually relieved by swallowing fluid, though the pressure may continue until the first bolus is forcibly ejected as a projectile vomit.

Treatment

At this time and whilst the sac which ultimately commences to form is quite small, *dilatation of the cricopharyngeal area* of the œsophagus will give relief of varying duration. Negus (1950) suggests the use of a hydrostatic bag for the dilatation. Also if chronic hypopharyngitis is present iron may be required and a powder such as barium sulphate together with orthocaine to protect any ulcertaion of the mucosa, to relieve discomfort and help relaxation, will be useful before meals.

As the sac increases in size and passes downwards behind the posterior œsophageal wall, forcible dilatation of the cricopharyngeal area becomes less effective in relieving the patient's symptoms and whilst the sac is an inch or so in diameter and before it is closely in contact with a comparatively large area of œsophageal wall, the most satisfactory treatment, if the symptoms warrant intervention, would seem to be *division of the circular cricopharyngeal* muscle fibres by the external route. This manœuvre should be followed by inversion of the sac by means of a purse-string suture, bringing together neighbouring tissues over the gap in the posterior pharyngeal well. Kaplan (1951) and Hoover (1955) have described division of the cricopharyngeus muscle for dysphagia due to poliomyelitis. At this stage excision of the sac itself is difficult and not as satisfactory as in the later stages. Usually, however, the symptoms at this stage are less marked than at an earlier or later stage and little accumulation of food occurs in the lumen of the sac. In practice it is not difficult to expose the cricopharyngeus muscle and to divide it without opening the pharynx. It is almost four years since I carried out this procedure for the first time and the patient is still fit and well with no recurrence of symptoms. Negus mentions the possibility of œsophageal respiration as a complication of division of the muscle, with ballooning of the œsophagus and elimination of peristalsis leading to cardiospasm. The muscle has been divided many times during division of the common wall between sac and œsophagus and no such complication has occurred in the experience of Professor Dohlman or myself, not have I found an account of it in the literature.

The *third stage* is when the difficulty in swallowing becomes constant and resolution is unlikely, the sac is large and a considerable residue of barium is constantly observed on X-ray examination. This is the stage when extirpation of the sac may be carried out with fewest complications and with a smaller chance of recurrence. Unfortunately, in these cases the patient is seldom under 50 years of age, often not a good operative risk, and his nutrition may be seriously defective as he is able to swallow only small amounts of food. Many of the patients are of nervous type and refuse all but the most minor surgical treatment, often entering the hospital in a seriously emaciated condition, hence it is frequently necessary to limit operative treatment to the minimum consistent with the relief of the dysphagia. If the sac does not appear to be adherent to the œsophageal wall at this stage, then removal by the external route, together with division of the cricopharyngeus is the best treatment (Fig. 1).

A large sac, closely adherent as most are, to the œsophagus, may be treated by division of the adjoining wall by endoscopic diathermy.

Mosher (1917) was one of the first to describe division of the septum between œsophagus and diverticulum, but he performed the operation with scissors and without the aid of diathermy or antibiotics. Mosher was led to try the operation when a case failed to respond to dilatation of the œsophageal opening by bougies. He described six cases (1929) in which he divided most of the septum without complications, but a later case of the same author's died of mediastinitis.

Dohlman (1949), who has used diathermy to divide the common wall in more than 80 cases with neither mediastinitis nor other serious complication, states that an external operation is unnecessary unless in view of other factors it is considered desirable to excise the sac. Endoscopic diathermy division of the opposing walls of the diverticular sac and the esophagus, including the cricopharyngeus muscle, will allow the contents f the sac to escape and lessen the muscular barrier to the passage of food eyond the upper œsophageal sphincter. Little disturbance of the patient ccurs and post-operative discomfort is very little. Dohlman considers the langer of mediastinitis is less than in the external operation for the emoval of the sac. Schubel (1947), Bayer (1948), Larsen (1954), Novotny 1953) and Bronner (1953) record experiences similar to those of Dohlman. Extension of the sac below the clavicle does not affect division of the ommon wall, though it may at times contra-indicate removal (Kulvin, 1944). It is likely that there is some adhesion between the opposing walls by the time the sac is large enough to cause serious symptoms, this tendency of the walls to adhere would be increased by diathermy coagulation, particularly if the current be passed *viâ* forceps grasping the ledge for a few seconds before division is completed with a diathermy needle.

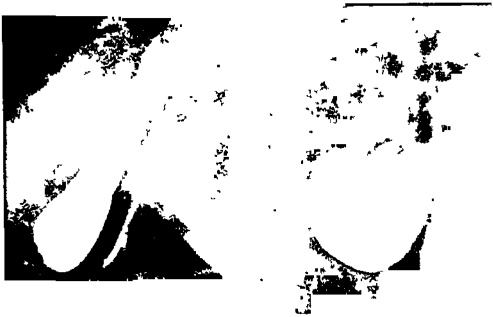


Fig 1

Lateral and A-P views of sac, showing space between sac and posterior oesophageal wall which may constitute a contraindication to endoscopic division of 'common' wall

Monkhouse (1957) has pointed out that it is difficult to divide the wall with forceps and diathermy as the coagulation resulting is not localized but becomes widespread along the wall of the œsophagus. The use of the divided ended œsophagoscope as the second electrode lessens the spread of the coagulation. It is usually unnecessary to do more than two stages, but occasionally some fibrosis in the divided area makes a further dilatation some months later advisable.

Details of Operation

Pre-Operatively: If the sac is large it is washed out the evening before operation and thereafter nothing is taken by mouth. The operation is carried out through a short wide œsophagoscope or Dohlman's œsophagoscope (Fig. 2) with a jaw-like slit in the distal end to fix the common wall and after it is located the ledge between diverticulum and œsophagus is seized in the midline with a pair of endoscopic crocodile forceps, the shaft of which may be partially insulated with thin waterproof adhesive tape and the diathermy endpiece applied to the proximal end of the forceps and the second electrode attached to the œsophagoscope, care being taken that nothing other than the mucosal ledge is touched by the forceps. Coagulation is continued for a few seconds and the forceps are removed, replaced by a long insulated wire to act as a diathermy needle and the first stage of

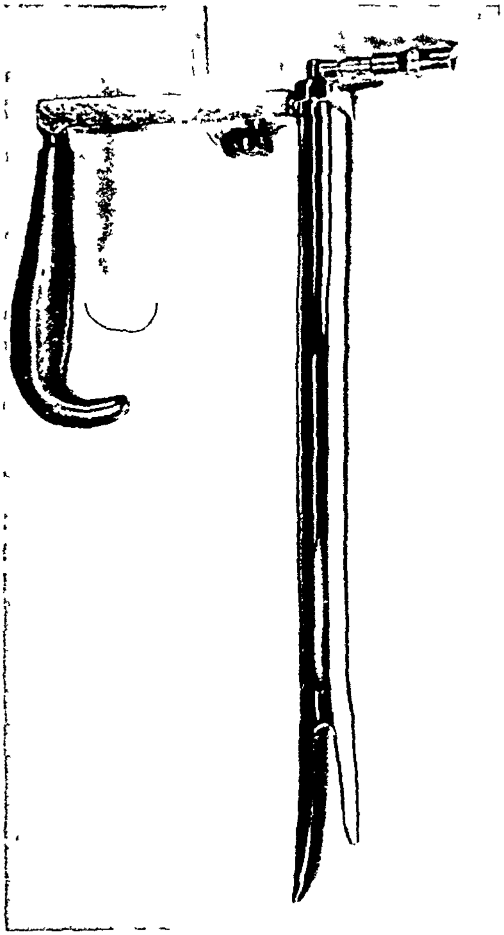


FIG 2

Dohlman's spatula-proximal lighting and divided distal end.

division is completed. The entire segment to be divided (up to three-quarters of the length of the wall) may be dealt with at one stage, but the operation is usually completed in two or more stages. It is probably not advisable to dilate the upper opening of the œsophagus before cutting the ledge as this procedure may disrupt some of the adhesions joining the two walls, but it is sometimes useful if a Dohlman type œsophagoscope is not available, to open the œsophageal mouth with a small gum elastic bougie to separate the walls whilst diathermy is being used. Dohlman states that he has not seen hæmorrhage during or after operation, but has designed a long needle with which to sew together the edges of the cut wall. This, or another form of needle, may be used to transfix and ligate the ledge instead of dividing it. The ligature if tied fairly tightly will produce an effect similar to the diathermy. Hæmorrhage might also be avoided by crushing the spur with long artery forceps which would be left in situ for a few minutes before the division. If more of the spur were crushed than was actually divided at the first stage adhesions would be ensured before the next operation. Bleeding has not occurred in the present series of cases. Scar tissue is seldom troublesome, but in smaller sacs it is occasionally necessary to repeat the diathermy division owing to scar contracture, though this latter appears to be evidence of too short a division of the ledge. Dohlman states that this contracture occurs in 7-8 per cent. of cases and has been much less trouble since the œsophagoscope was used as the second electrode. The present advanced state of anæsthetics makes local anæsthesia rarely necessary, but if this is used it may be advisable to infiltrate the ledge with a small quantity of procaine and adrenaline before applying the diathermy. Legler (1952), who has treated many cases of pharyngeal diverticulum in this way, and Dennhardt (1950), both carry the incision down to a point below the fundus of the sac to open the œsophagus as widely as possible. *Post-Operatively* the patient should have sterile fluids by mouth for 36 hours with 250 mgm. tetracycline as an emulsion two-hourly for the first 12 hours and six-hourly for another day, when he may usually leave hospital.

The possibility of the subsequent development of cancer in the sac of a diverticulum after the endoscopic operation has been raised by Lodge (1949), but no record of such a case has been discovered in the literature though several articles have appeared describing the discovery of a carcinoma in the pharyngeal pouch (Socin 1916, Seiffert 1927, Lotheissen 1930, Capps 1933, Sparks 1933, Konjetzny 1934, and Cabot 1940), but in almost all of these the diverticulum has been of long standing and the stagnation marked. In other cases the tumour has been present before what appeared to be a diverticulum was seen (Edgren and Quensel 1890, Keppler and Erkes 1919, Seiffert 1953). The development of a carcinoma at the site of a diverticulum previously removed by the external approach has been described by Kelson (1921). Long-standing stagnation of food in the sac may thus predispose to the intra-diverticular neoplasm, but the operation which divides the common wall and the spastic muscle fibres responsible for the hold-up must, to a large extent, relieve the stasis and considerably lessen the chances of the development of what is in any case a particularly rare lesion.

As far as *contra-indications* are concerned diathermy division of the common wall of sac and œsophagus appears to be effective only in cases of pressure diverticula herniating above the contracted circular fibres of the cricopharyngeus muscle. A sac may form secondarily to web formation in the upper œsophagus (Mosher 1917, and Seiffert 1953) in certain cases below the level of the circular muscle fibres. In this case division of the common wall will usually fail to relieve the symptoms if the stricture is more than 2 to 3 centimetres in length and increasing dysphagia may result as further fibrosis occurs. Hence this operation appears to be contra-indicated when a fibrous narrowing of the upper œsophagus is present.

Some of the continental authorities (Legler 1954) state the results in the very large sacs are not always good, but this may be due to marked hypertrophy or even calcium deposits in the cricopharyngeus muscle which may then be very difficult to divide. If the symptoms recur it is simple to carry out another stage of dilatation or division of the common wall. If after external removal recurrences take place as has happened in 10 per cent. of some of the large series, re-operation of any type is difficult.

It is interesting to review some of the cases treated by one or other of these methods.

Miss E. B.

It is sometimes very difficult to foretell on X-ray and clinical findings what treatment is advisable and endoscopic examination should be carried out in all cases (Figs. 3 and 4).

In this patient, an old lady of 77 years, complaining of severe dysphagia and with a hæmoglobin of 50 per cent. all that was required was dilatation of the œsophageal opening. This was done a year ago and the patient has had no further trouble.

Mr. A. F., æt. 70 years.

Patient referred with dysphagia, cough and loss of weight.

14.12.53. Direct examination under anæsthetic showed a small sac with considerable narrowing of the œsophageal opening. This was dilated to admit the largest œsophagoscope.

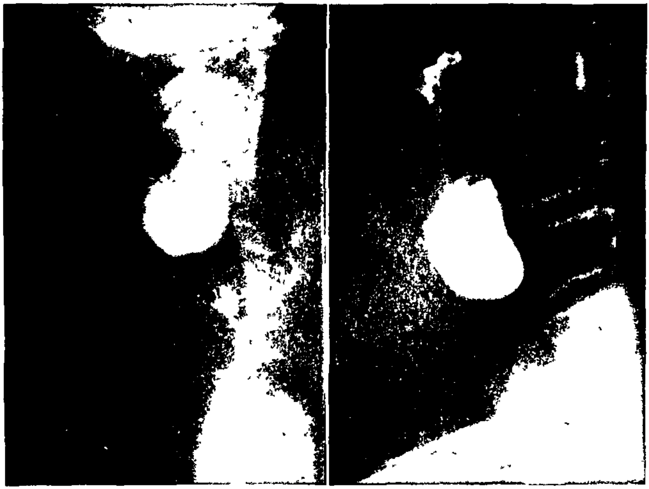
29.1.54. As condition of patient was not improved by dilatation the sac was exposed through an incision in front of the sternomastoid and lifted upwards to demonstrate the circular muscular bundle of the crico-pharyngeus which was divided with scissors. The sac was inverted and a double purse-string suture inserted to draw the weak area of the pharyngeal wall together.

24.2.54. Patient was now swallowing without difficulty.

At post-mortem three years later, after death from an intra-abdominal condition, the diverticular sac was found to be represented by a small fibrous nodule inside the lumen on the posterior wall of the œsophagus.

Mr. A. Br., æt. 52 years (Fig. 5*a*)

Symptoms of short duration, but severe. Loss of weight, regurgitation, cough and dysphagia. This patient was found on œsophagoscopy to have a sac 1½ inches in length.



FIGS 3 & 4.

Diverticulum in lady of 77 years Condition responded to dilatation of oesophageal opening

Lateral view of Fig 3

It is unusual to find that only one stage of division is required, but in this case the sac contracted (Fig. 5*b*) after the first stage and the swallowing and general condition of the patient improved rapidly.

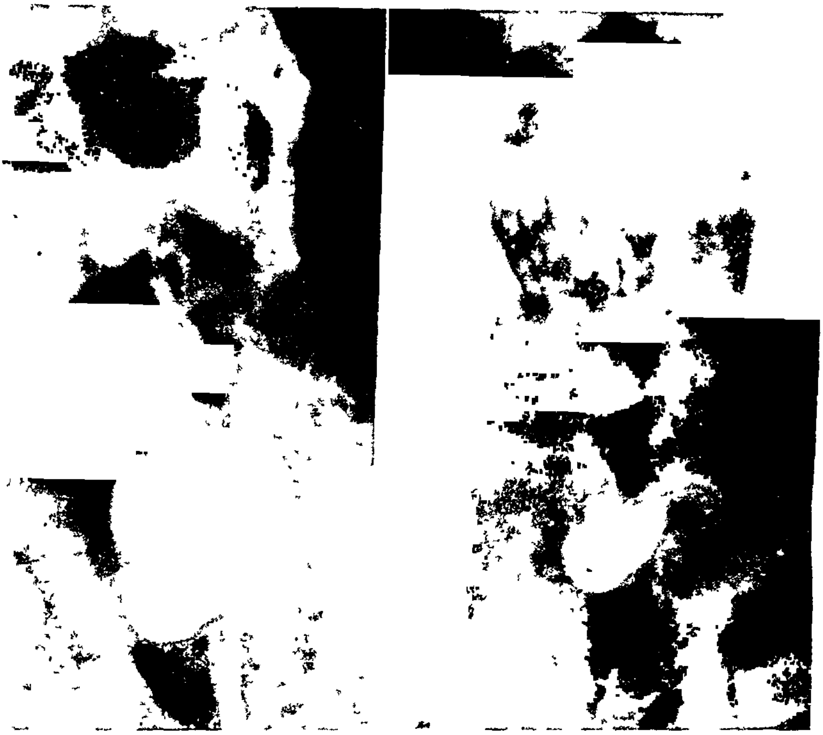
Mr. A. B., æt. 74 years.

First seen March, 1949, complaining of upper dysphagia of one year's duration and loss of weight. Barium swallow showed a very large pharyngeal diverticulum, but patient refused to have the sac excised.

In May, 1949, patient was admitted with almost complete œsophageal obstruction, very dehydrated, ill and very emaciated.

26.5.49. Gastrostomy carried out by Mr. G. A. B. Walters under local anæsthesia. After this, patient's condition improved, but he did not appear fit for the operation of excision of the sac and it was decided to divide the common wall with diathermy.

26.7.49. Operation was carried out through a short œsophagoscope. The upper œsophageal opening was difficult to discover, but was finally found and the ledge divided for three-quarters of an inch.



FIGS 5a & 5b

X-ray before operation.

X-ray after one stage division of common wall endoscopically shows considerable reduction in size of sac

2.8.49. The second stage of the operation was completed with no difficulty. Thereafter the patient's condition improved rapidly and he stated that he had no further difficulty in swallowing.

February, 1950. Patient's condition was excellent and he was 5 stones heavier than in May 1949. A barium swallow since operation had shown no real change in the size or appearance of the sac, but as the patient was obviously fit and well and had no dysphagia, further division of the party wall was not indicated. The improvement appeared to have resulted from the division of the cricopharyngeal sphincter and the complication of air swallowing in the absence of some protecting mechanism in the post-cricoid region did not occur.

July, 1954. Patient's condition remained satisfactory.

April, 1955. Patient was admitted with strangulated inguinal hernia and died shortly after operation. Specimen (Fig. 6) consisting of larynx, pharynx, upper trachea and œsophagus, shows the diverticular sac and a wide œsophageal opening considerably farther than normal from the arytenoids, the result of division of the common wall.

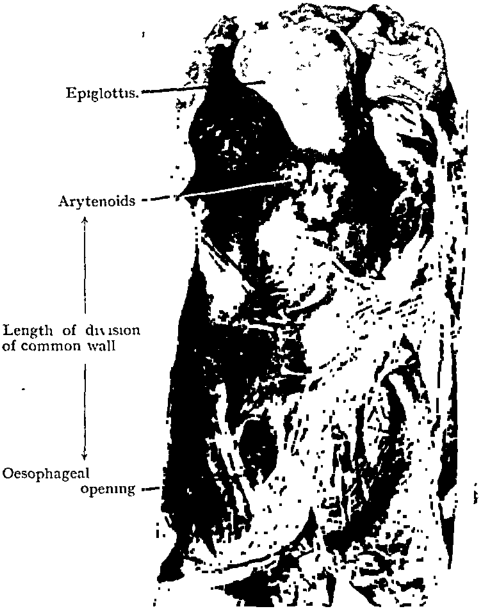


FIG. 6.

Specimen of larynx, œsophagus and pharynx removed 6 years after division of common wall.

Mr. J. T., æt. 70 years.

Patient was first seen in 1947 complaining of dysphagia and regurgitation of food with loss of weight and was found to have a large pharyngeal pouch, but refused to have treatment for the condition.

He next reported on 22.9.52. The pouch had enlarged and could be seen bulging on the left side of the neck. On this occasion the patient consented to endoscopic treatment of the pouch. The œsophageal opening was found with some difficulty high up on the anterior wall of the sac and the dividing wall coagulated for an inch or so with diathermy. Two further stages of the division of the wall were completed within the month, the patient returning home within two days of each stage. In early December the patient reported very much better and he had gained a stone in weight. His condition remains very satisfactory, but the X-ray appearances have shown little change.

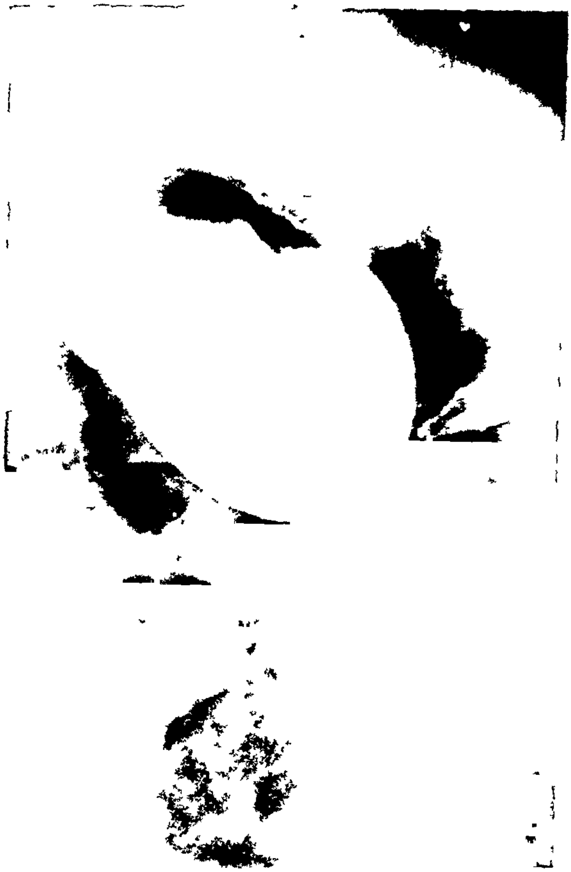


FIG 7

Very large sac in patient of 86 years treated successfully by endoscopic means

Mr. C. E. I., æt. 86 years (Fig. 7).

This patient commenced with mild dysphagia shortly after the first world war. At first no sac was demonstrable radiologically. The dysphagia had increased considerably and the patient found it necessary to bring up food between the courses so that his weight was 7 stone 6 lb. (height 5 feet 9 inches) Regurgitation into the nose was unpleasant and embarrassing The patient had myocarditis and heart block and his general condition was poor

Barium swallow only a dribble of barium got away from the sac after over half a pint of barium had filled it, regardless of posture, etc Prominence of the interstitial markings in the right lower lobe suggesting fibrosis ~~as~~ a sequel to aspiration pneumonia

His hæmoglobin was 65 per cent, red cells 3,000,000, the result of starvation, corrected with two pints of blood

13 2 55 First stage of division of the common wall by diathermy. Huge diverticulum discovered First and second inches of the œsophagus found to be markedly narrowed As it was unlikely that the first stage of division in a sac of this size would relieve the dysphagia sufficiently to improve the patient's nutrition, a stomach tube was passed The cough became worse as a result of this tube increasing the tendency to overflow into the larynx

22 2 55 Second stage One and a half inches of the ledge divided A firm, almost cartilaginous, area of muscle was encountered, which was difficult at first to divide with diathermy

25 2 55 Swallowing was now almost normal and patient took an ordinary diet

2 3 55 Weight 8 st and condition much improved

25 7 55 Weight 8 st 10 lb Condition continued to improve

15 11 55 Weight over 9 st General condition good

In no case has any notable infection of the surrounding tissues occurred, though a little tenderness of the neck has been present at times for a day or so Neither air swallowing nor regurgitation of stomach contents have been encountered It has been necessary in one case to dilate the upper œsophagus owing to fibrosis and contracture of the scar

In two cases the division has not been completed In one case the patient decided to have the sac removed instead of the second stage of division of the common wall The external operation was not, I understand, made more difficult by the endoscopic operation In the second case I was unable to divide the very hypertrophied cricopharyngeus muscle in two stages and advised a further stage which has not as yet been done

Thus the methods advocated for treatment of pharyngeal pouch

Early—(1) Dilatation of cricopharyngeal sphincter endoscopically only 2 cases

(2) External division of cricopharyngeus and inversion of sac 3 cases

(3) Later, but before sac adherent to œsophagus—removal of sac and division of muscle. 5 cases.

(4) Adherent sac—diathermy division, particularly in patients over retiring age. 17 cases.

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RECENT ADVANCES IN TYMPANOPLASTIC SURGERY*

By F McGUCKIN (Newcastle on Tyne)

Introduction

ONE HUNDRED AND TWENTY years ago a distinguished professional forbear established the first otological hospital in the world, The Metropolitan Ear Institution, now The Metropolitan Ear, Nose and Throat Hospital According to Wilfred Trotter it is right and proper that we should commemorate the lives and works of celebrated men, and so today we honour James Yearsley, who founded the Metropolitan when he was 33 years old A native of the West Country himself, he might have approved of a provincial lecturer, after a string of metropolitans, two from the English Metropolis and one from the Scottish Because of his interest in artificial ear-drums he would certainly have agreed on the subject selected for this occasion by the Dean and Medical Staff Committee of his hospital Moreover, Yearsley was an advocate of the Eustachian catheter and, had he been living today, you might easily picture his delight in the recent experiments with indwelling tubes for the further salvage of the less hopeful cases of destructive ear disease Whether he would have agreed the choice of person for this lecture is less certain, because like repels like, and James Yearsley was noted for his intolerance and impatience

Evolution

This predecessor of ours told his own tale, a tale based on personal experience It has been my good fortune to watch the evolution of what we now call tympanoplastic surgery during the past decade and I shall follow the example of James Yearsley by unfolding a story based chiefly on observation and discussion, rather than on an arid survey of the literature What I have to say derives directly or indirectly from Fraser, Tumarkin, Thorburn, Marshall, Livingstone and Beales in Britain, from Lempert and Juers in America, from Verhoeven and Groen in the Low Countries, from Pietrantoni and Bocca in Italy, and most of all from

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* The Fourth James Yearsley Memorial Lecture given at the Royal Society of Medicine March 7th 1958 The lecture proper was based on a large number of illustrations so that the published script forms little more than a summary

Zöllner, Wullstein, Kley and Shicker in Germany. And it is worth noting that some of these people have written little, yet contributed much.

Just after the last war, I made time to look at some of the local results of my chronic ear surgery. As McEwen would have said, the picture was not pleasing to us. Visits to Tumarkin, Thorburn and Otty gave rise to discussion and disputation, and also to ideas—the concept of accessible and inaccessible disease, of reversible and irreversible pathology, of the distinction between the term radical in relation to disease and radical in relation to function, of excisive surgery as opposed to radical surgery so as still to leave room for separate discussion of function, and so on. These concepts were later and quite separately to be clarified, codified, and applied by continental surgeons.

Late in all this argument, which coincided with the more general use of the operating microscope, I found myself able to appreciate why some "radical" mastoidectomies retained useful hearing. A patient of mine had been submitted to a radical operation by Milligan in 1929. He undoubtedly had useful hearing although the other ear was destroyed by cholesteatoma. Repeated scrutiny suggested that there was an air-containing hypotympanic tunnel, shaped like a sausage, and covered by a just visible sheet of epithelium generated from the inferior annulus. I christened this little tunnel a round-window baffle, and afterwards some of my less polite friends called it the McGuckin sausage. Here was my first introduction to accidental tympanoplastic surgery, though the name tympanoplasty was as yet unborn. One case, however, was far from fulfilling Aristotle's law of sufficient enumeration; yet after all, the situation was much like that of a standard Lempert fenestration, one window inside the tympanum and one window outside. Subsequently I was able to recognize other mechanisms conducive to good hearing, most of them arising from natural healing *and many having no history of ear disease.*

In 1951, Louis Verhoeven wrote telling me of a radical operation with remarkable hearing. The prediction of a round-window baffle was agreed later when we examined the case together in Antwerp. Thereafter we tried to reproduce these accidental results by tacking remnants of tympanic membrane to the promontory below the oval window. My own success was so slight that as late as 1952 I was still preaching that disruption of the ossicular chain precluded functional repair. I followed this up by trying to convert some of these simple baffles into what Juers (1954), at the suggestion of Tschiassing, called myringostapediopexies. My title was even more unfortunate—myringodermostapediopexy. I say unfortunate not only because it was inelegant, but because we had in mind a mobile, not a fixed stapes. Zöllner was already far ahead, for he had previously described the situation as a columella-effect.

At this point I should make it clear that the validity of certain propositions will be *assumed.* (1) In the normal ear, the oval window is physiologically outside the tympanum, though anatomically it is inside (2) There is a lever system consisting of drum-head and ossicular chain, and through this a small movement over a large area (the drum-head) is transformed into a large movement over a small area (the oval window), giving a concentration of force at this point This concentration has been estimated by various physicists as being worth 15 to 1, 28 to 1, even 40 to 1, but the ordinary clinician must take his choice (3) In opposition to the impact at the oval window, there is sound-protection of the round window, arising in part from reflection of energy by the stiffness of the tympanic membrane, and in part from the cushion effect of the air space inside the tympanum (4) Sound protection of the round window alone is sufficient to give adequate speech hearing provided the oval window is mobile and not physiologically inside the tympanum (5) There should be only one *mobile* window on either side of the basilar membrane

These propositions and the "equations arising from them were shown diagrammatically (McGuckin, 1955) in the discussion which followed the opening papers of Zollner (1955) and Pietrantoni and Bocca (1955) at the Royal Society of Medicine in May, 1955

In 1953 there were some who, like myself, arrived in Amsterdam unaware of the work published in the previous three years by Moritz by Zollner, by Wullstein, and by Antoli Candela My ignorance of the German and Spanish languages alone precluded foreknowledge Consequently I was not alone in receiving a rude shock when Zollner of Freiburg, and Wullstein, then resident in Siegen, launched their ideas of reconstructive ear surgery at the International Congress My first reaction was one of doubt, even disbelief, but private discussion with these men—and how tolerant they were—soon convinced me of their real progress The significance of the work lay in the bold handling of free skin, the imaginative preparation of suitable bedding to nourish the free-skin, the meticulous stripping of the outer cuticle of the drum head, the visual control of movement in both windows, the use of amnion to encourage mucosal regeneration beneath it, the early maintenance of the new tympanic cavity by gelatin beneath the graft, and so on This was a new world wherein techniques were already being rationalized, as the illustrations in the Congress Proceedings show

I well remember my discussion with Wullstein He asserted that we must have mucous membrane from somewhere, by regeneration or by other means, and as you know he has since employed buccal mucosa as a substitute, with success at times Thorburn (1954) was obviously thinking on similar lines when in a review of 188 cases of radical and modified radical operations he demonstrated that he was trying to integrate the older techniques with the newer It is now four years since he sounded a note of caution, particularly about covering up disease, and in my view the warning remains as valid today as it was then

Meanwhile my equations were in process of difficult labour and Wullstein was occupying himself to much better purpose. In the bitter winter of early 1955, Verhoeven, Marshall, Thorburn and myself went off to Germany, all agreed on what we were going to see. Wullstein had by now constructed the scheme of his five types. My friends were quick to point out that two of my seven possibilities which varied from complete reconstruction to complete destruction of function, were not tympanoplastic at all, and I was only too pleased to settle for Wullstein's formula —though still having some reservations arising from our visit to Zöllner.

In my view this was really the crucial moment. In three years, largely because of the work of these two Germans, a few exploratory ventures had been converted into a planned campaign. The use of whole skin, and sometimes of split skin, was established as a useful method of reconstructive ear surgery. It was known that whole skin must be so thinned down that its plasma spaces might share easily with the plasma spaces of host-tissue; that the bed must be prepared in a fashion to nourish the graft and, eventually provide a blood supply; that to do this it might be necessary, according to circumstances, to strip the outer cuticle of the drum-head and the skin of the adjacent auditory canal. The principles of excision of irreversible disease and of adequate cochlear function were agreed, but there remained then—and even now—many elements of uncertainty, e.g. the difficulty of the recognition of irreversible pathology, doubts about the use of the artificial columella, about the total excision of cholesteatoma matrix, about the behaviour of skin in the mastoid segment, about the positive virtues of the acoustic probe, and even about the place of myringoplasty itself in the scheme of things.

Types

And now it is appropriate that we look at the five types of Wullstein and consider the problems which they present, and also the variations which have come from experience. The composite diagram has been used in Britain several times and is now well known; it has been employed by Thorburn more than once, most recently in the *British Medical Journal* (Thorburn, 1958), and there is no need to reproduce it again.

Type I for a good result requires normal sound-transformation at the oval window and normal sound protection at the round window. These are conditions which may be fulfilled by a simple myringoplasty, if the perforation is not large and if the disease is genuinely inactive. Wullstein has almost no place for myringoplasty as a separate entity, saving only the case of trauma, whether accidental or surgical. In his view any technique should make three "controls" available, namely control of the antrum, control of the attic, and control of the tympanum, this last to include particularly the control of both windows. The technique is summarized in a beautiful picture taken by Walter Kley and published by Roche (*Medizinischer Bild-Dienst*, April, 1957), demonstrating full inspection of the antrum, of the attic above an attenuated bridge and of the tympanum below the bridge viewed through a Wullstein burr-hole driven between the corda tympani and the facial canal

Now in a simple permeatal myringoplasty, the first and second controls are not possible, and the third control is doubtful Zollner does have a place for simple myringoplasty but, if I understand him aright, he clearly requires a fair certainty of the absence of hidden disease Philip Beales (1957) and Stuart Mawson (1958) in this country have discussed their experience of myringoplasty, and the results may not be dismissed lightly Livingstone (1958) on the other hand thinks we should abandon myringoplasty as a term, or that at best it should be regarded as one possible variant of type I tympanoplasty The discrepancy between the two positions cannot be wholly resolved Wullstein does allow that, in given conditions, not all of his three controls are necessary, but he does claim that they should be available, and he goes on to say that myringoplasty, with Thiersch grafting, was tried 70 years ago by Berthold and Tangemann, but it was abandoned (Wullstein, 1956) My position is similar to that of Livingstone, but it may be because of the kind of material which occupies so much of my operating time Nevertheless it is surprising how often a lesion which looks inactive does prove to be otherwise when the cleft is inspected Having paid two very recent visits to Germany for conversations, I would quote from a letter written to me by Wullstein on January 21st, 1958 "In the German otology, tympanon means the whole content of the middle ear and the idea was to express that the whole tympanon with the hearing bones, the windows and so on, the epitympanon and the end of the tube had to be controlled So I was convinced that tympanoplastie would be the name to express that these operations could only be done if the whole middle ear was controlled and rebuilt That is why I cannot agree with the name myringoplastie Myringoplastie means only the reconstruction of the tympanic membrane It is an old name used already by Berthold and Tangemann in 1880 in Germany and reintroduced by Zollner, but a myringoplastie is only the last part of tympanoplastie" He then goes on to give his reasons for the decision, saving only the case of trauma (Wullstein 1958)

I had planned to talk with Zollner only two months ago but his illness made the visit impossible and on my part I could not make a second journey to Germany within a month Not having had personal contact with him for three years now, I must fall back on the literature and correspondence He undoubtedly has a place for permeatal myringoplasty but he has also recently discussed an operation called meatantrotomy (Zollner, 1957) This technique is used when the meatus is too narrow for easy transmeatal closure of a dry central perforation, or when the pathology demands inspection of the antrum and the windows. It really implies a method of providing antrum control and window control. Pleasing results have been attained, but Zöllner remarks that he would be happier if there were more cases suited either to simple plastic closure or to meatoantrotomy. It may be said that these two continental proponents have moved very slightly towards a middle way, but some discrepancy remains. Having respect and admiration for both, I would myself come down on the side of Wullstein and Livingstone by saying that simple myringoplasty has at best a very limited place.

In the life of any individual surgeon, the proportion of cases falling to types I and II must vary with the kind of material presented to him and the time he must devote to less promising stuff. Verhoeven, whose tympanoplastic experience is rivalled only by the Masters themselves, has the finest collection of colour photographs I have seen anywhere and from two or three hundred of these I was privileged to take my pick. They demonstrate all five types and include three magnificent examples of type V, two showing tympanosclerosis. Thorburn's case represents a successful type III carried out for traumatic disruption of the ossicular chain (Thorburn, 1957). Verhoeven's pictures are unpublished and present-day costs would make publication prohibitive.

Difficulties

And now it may be well to turn to the difficulties and disappointments which have attended this immense advance in chronic ear surgery, or at least to consider some of them. In 1952 I surveyed the case for the patient as a whole and in principle there is little reason to vary the approach then laid down. The owner of the discharging ear or of the large perforation is more important than the local condition, and the advice we tender about one ear must often depend on the state of the other ear. The age and condition of the whole patient also demand consideration. What concerned me in 1952 was the production of healed ears, and it is still a matter of concern now, if one is dealing with long-standing continuous otorrhœa, or with cases about to submit to a third or perhaps an eighth operation.

I do not agree with the theory that all may have tympanoplasty, if by the term we mean reconstructive functional surgery, because there are limits to the possibilities of surgery. I have had some gratifying tympanoplastic results varying from type I to type IV and some of my own photographs confirm this.

Last year at the Bristol meeting (McGuckin, 1958) I surveyed a series of 70 consecutive cases, later brought up to 82. The final script was lost in the B.M.A. Annual Meeting in 1957, but my notes indicate that of the 82 cases, 19 already suffered from invasion of the labyrinth (not just a fistula), 23 showed pathological exposure of the facial nerve, 70 had frank cholesteatoma, and in 77 the ossicular chain was disrupted. This was not very promising material, since in nearly a quarter the potential of functional salvage was nil from the very start. In this group a sequence of operative photographs in one case begins with cholesteatoma everywhere and ends with the facial nerve crossing the temporal bone in splendid isolation—and incidentally still having splendid motor function. Another picture illustrates a huge mass of keratosis which had destroyed everything except the facial nerve.

Two years ago, four groups of cases were presented at the North of England Otological meeting in Newcastle. They were designed to show that results must be related to material, and also to show how far surgical ambition had succeeded or failed. In the first group were patients in whom the full potential had been attained, for example a complete reconstruction (type I), or a satisfactory round-window baffle (type IV). In the second group cases were presented in which function fell short of that desired but in which there was some gain—e.g. an attempt at columella hearing (type III) had given a useful baffle (type IV). The third group was composed of cases wherein functional salvage had failed completely, and in some of these foul discharge persisted. The fourth group was made up of seven cases in which the potential of functional salvage was nil or nearly so, and it included several examples of labyrinthine invasion. Some of these seven cases I would now regard as less hopeless because I believe that Beales is right in saying that by delaying the completion of an operation for one, two or three weeks, regeneration of inner-wall mucosa may provide a more favourable basis for reconstruction.

Twelve months later Thorburn, also at a North of England meeting held in Blackpool (Societies Proceedings, 1957) showed a variety of tympanoplastic results of all types, including eighteen cases in type III, and in so doing demonstrated how much more could be done with better material and, if I may say it, with greater skill.

This meeting produced a lively discussion, centred round difficulties and disappointments, rather than successes. Thorburn drew attention to the frequent failure of whole-skin when used to line the mastoid segment, and mentioned that he had tried to improve results, both in tympanoplasty and in the Wullstein free-graft type of fenestration, by cutting up part of the skin and applying it as a "crazy paving" in the mastoid region. Beales showed histologically that a large sheet of skin might survive well in the tympanic and epitympanic regions, but that in the mastoid region it lost its specialized structure and became atrophic. Subsequently he has pursued a new method, not yet published, worked out in conjunction with Wilfred Hynes, a plastic surgeon. (Beales was himself trained as a plastic surgeon.) Their aim is to implant a living flap into the mastoid area, and to supplement this by using whole skin from the arm for the tympanoplastic reconstruction. The gap behind the ear is covered by split-skin and so far healing has been encouraging. I am privileged to show you pictures which illustrate this new approach and am grateful to these two colleagues for their kindness. R. M. Marshall and J. Potter are at the present time conducting rather similar experiments and they have obtained some pleasing results. These two teams, each consisting of an otologist and a plastic surgeon, must presently give all of us something to think about.

Some of us had the opportunity of discussing the problem with Wullstein six months ago. There is no doubt that he has had difficulty and he suggested that perhaps tight packing was a factor which might set up necrosis in poorly nourished skin, two to four weeks after operation. With only a limited experience since then, I have the impression that gentle moulding with moist gelatine, surmounted by a very light pack may be helpful.

The debate at the North of England meeting then turned to the cases of type III at which point Thorburn stated certain principles. The surgeon, having decided the merits of the individual problem, and agreement being reached to proceed to operation, he must then:

(*a*) Assess and eliminate irreversible pathology, e.g. osteitis, granulations or keratinizing epithelium, from the tympanic cavity and the mastoid.

(*b*) Assess and respect reversible mucosal pathology so that a mucous membrane is retained over the medial tympanic wall.

(*c*) Test the functional integrity of the ossicles and (? *also or*) of the oval and round windows.

(*d*) Prepare a bed for the full thickness graft used to create a new drum by stripping away the outer epithelial layer from the remains of the drum and the skin from the adjacent meatal wall.

(*e*) Achieve a relatively bloodless field.

Beales then went on to refer to cases where extensive excision of disease was necessary and said there was a place for a two-stage operation. Under antibiotic treatment mucosa could regenerate rapidly, and a case which initially appeared hopeless might be safely and successfully concluded in two or three weeks.

Now I have to revert to the warning given by Thorburn in 1954. In my view complete excision of irreversible disease is necessary (I admit that opinion on this point is fallible), and irreversible disease for me includes every scrap of cholesteatoma matrix. It is true that Wullstein is willing to leave a small oval of matrix on an exposed stapedial footplate in type IV, but I am unwilling to go thus far. In my view matrix is disease, not useful epithelium, and I have for years supported Theo. Walsh in his campaign for its removal in surgical procedures. The most difficult problem which cholesteatoma presents to me occurs when we find nests of cells in the solid angle, and still more in the hypotympanic and peritubal areas, and when each cell is lined with matrix. Despite Tumarkin's opinion, these things do occur, and in my practice they occur often. It requires skill and courage to clear the solid angle, and it sometimes requires far more than my skill to be sure that cells in the hypotympanic and the peritubal regions have been cleared of matrix. Even the two-stage operation gives me no more than a glimmer of hope in such cases.

The battle cry of "tympanoplasty for all" reminds me of the little girl I showed at the B.M.A. Annual Meeting last July. She has survived lateral sinus thrombosis, meningitis, and three cerebellar abscesses on one side, in which the ear has been destroyed from the mastoid to the Eustachian tube. There is a fairly safe though suppurating cavity on this side and the other ear shows precisely the same cholesteatomatous mess—and it remains to be treated sometime. There is small hope of functional reconstruction here.

Marshall, Verhoeven, Thorburn and myself have observed a case operated on by a highly competent and skilful surgeon, in which there were three collections of cholesteatoma under a tympanoplastic flap two years after the intervention.

There must inevitably be disappointment and frustration if we deal with long-standing disease, but this does not mean that I deny the very real progress of the last five years. Nearly two years ago Terence Cawthorne came to visit me. He seemed pleased with what he saw, but I gave him a somewhat gloomy picture, chiefly because two days before I had an out-patient morning of the most depressing kind, when all my fœtid failures presented at once. Two days later, after a morning spent looking at a series of brilliant successes, I wrote to say that modern tympanoplastic surgery was not so bad after all.

It is beyond the wit of any otologist to cover the whole of this field in one lecture but, before proceeding to my peroration, there are other matters worthy of consideration.

It is relatively easy to produce good results in cases of inactive disease, much more difficult where pathology is active, and particularly when it has been continuously active for many years. Before undertaking surgery it is therefore one's duty to try to reduce activity as far as possible, by topical and systemic treatment, if necessary under in-patient observation. Such a precaution will reduce if not eliminate disappointment both for patient and for surgeon, and we may go further by considering the circumstances in which a slightly delayed two-stage procedure may be worthwhile. Moreover, some of our hopeless problems in which we have reverted to the classical radical will later proceed to give satisfactory spontaneous functional results by producing their own type IV conditions.

Attempts have been made, apart from provisional guesses about the best potential of salvage, to predict post-operative results on a measured scientific basis. This brings me to Zöllner's ingenious method of pre-operative prediction—the "Schallsonde", or as it is known in this country, the acoustic probe. With all my respect for Zöllner, I cannot accept this investigation as valid. Beales (1957) says that it is tedious to perform and rather unpleasant for the patient. He then describes a functioning chain in which the efficiency of the incus depended on a stapedial attachment by granulation tissue only. I am reminded of a case shown by Verhoeven to Marshall, Thorburn and myself in which the ear to be examined showed only something like a five to ten decibel loss. Without collusion, each of us concluded that a ball of cholesteatoma was acting as a very efficient incus—simply because each in his own experience was aware of such a mechanism.

Present day methods have exposed some of the claims made for the endaural approach to the tympanic cavity. For years I was a most enthusiastic advocate of the endaural technique, even to the point of making a colour-film of what I was pleased to claim as my own plastic method. But it is now seven years since I said that the only true merit of the technique was that it then gave me the best and easiest plastic flaps. In saying this I do not wish to detract from the debt we all, everyone of us, owe to Julius Lempert. In today's context, the endaural method has no special merit except in one circumstance. It is useful for a first-stage procedure in the exploration of a chronic ear problem, where there is grave doubt about primary reconstruction, because it leaves the postaural skin for another day. I do not believe, and never did believe, that it provides an entirely fresh or even a different view of the middle ear as opposed to what may be seen by a post-aural approach. May Julius Lempert forgive me for saying so!

My last detail relates to the attempts which have been made to get better results in type IV cases, where we have a small narrow tympanum and considerable risk of adhesions. The objective in type IV is to maintain a round-window cover in communication with a competent Eustachian tube. I have mentioned one method—the use of absorbable gelatin and it often succeeds. Zöllner (1955) described another method at one of our section meetings and illustrated the idea with a diagram showing a thread which was packed into the hypotympanum and led out through the nose *viâ* the Eustachian tube. The thread was removed after 8 to 10 days. During last year Wullstein developed this idea by employing a fine indwelling catheter with one end showing just within the tympanum and the other brought out through the nose. Inside the catheter there is a fine nylon thread which is carried round the hypotympanum to the niche of the foramen rotundum. The thread is removed in 7 to 10 days and then controlled air-inflation is carried out daily by using an ordinary syringe with a small needle just fitting the catheter. Half a millilitre or perhaps two millilitres may be pumped in according to requirements. The air, however, does not always take the direction desired by the surgeon, and the technique is still very much under trial.

Peroration

And now to the peroration. Doubtless the origin of modern tympanoplastic surgery came from the accidental observations of those intelligent enough to see, but happily some observers pursued their findings with greater skill and better logic than did others. The seeds of this new plant were sown in scattered fashion less than ten years ago, and within the last five years we have been fortunate enough to see the full flower. What a glorious decade it has been in a sphere of surgery which, according to the prophets of gloom, has been dying or dead for these twenty years or more. It is clear that a lot has been learned, and there is still much to learn, particularly about the prevention of that pathology which causes all this pother. However, I take the view that by every test, Wullstein and Zöllner emerge as the fathers of our present-day plastic methods, as the fathers of this enormous advance in less than a decade, and I say that they are worthy of honourable mention on this day when we commemorate the life and work of the celebrated James Yearsley.

The Dean and the Medical Staff of the Metropolitan Ear, Nose and Throat Hospital invited me to deliver this lecture, the fourth in the series, but the honour—and it is a great honour—really belongs to the other three full members of the Cholesteatoma Club, Verhoeven of Antwerp, Marshall of Middlesbrough and Thorburn of Blackpool.

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GIANT-CELL EPULIS

ANALYSIS OF 26 CASES

By HENNING SØRENSEN (Aarhus, Denmark)*

Introduction

THE ætiology of giant-cell epulis has been the subject of much discussion; it is still obscure whether it is a neoplasm or a lesion in which trauma is an inciting factor, and its relation to giant-cell tumours of the long bones is another controversial point. It is possible that several types of giant-cell epulis differing in origin exist.

In order, if possible, to contribute to the elucidation of these problems, all cases of giant-cell epulis treated in the Radium Centre for Jutland and Aarhus Kommunehospital since 1935 have been reviewed and the results of treatment analysed. The results of this analysis show that *in most of these cases the epulides must be considered to be neoplasms, and that traumatic injury cannot be demonstrated to have played a part as an inciting factor, but may have accelerated the growth of the tumours.*

Historical Review

The term *epulis* is of Greek origin, meaning "upon the gum", and thus actually designates any outgrowth from the gingiva, whether fibrous, angiomatous, inflammatory or a giant-cell tumour. However, a distinction is usually made between two forms of epulis, viz. *fibrous* and *giant-cell epulis* (Boyd). In Denmark, *the designation epulis is used in the histological diagnosis only when the tumour contains giant cells*, and it is this form which is considered below. It is usually regarded as a variant of the giant-cell tumours occurring in the long bones. The ætiology of these tumours is not fully clarified either.

The designation giant-cell tumour was introduced by Bloodgood in 1910. He maintained that this tumour was benign, and the idea of benignity gradually gained ground and superseded the view which had been held in the nineteenth century, viz, that it was malignant, or that it might at least "degenerate" into malignancy Before Bloodgood's introduction of the new term, the growth was usually described as a myeloid tumour or myeloid sarcoma Thus, while it is now generally agreed that the tumour is benign, considerable difference of opinion as to its ætiology still exists, some investigators postulating that it is referable to traumatic injury, others that it is a genuine tumour

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Geschickter and Copeland (1930) strongly emphasized that trauma was an important factor in the development of giant-cell tumours In support of this view they gave the following reasons

Trauma may block the periosteal arteries for example, by the occurrence of a hæmatoma The osteoclasts are activated to provide new canals The impaired blood supply results in necrosis of the bone tissue from which the osteoblasts are supposed to form new bone The normal equilibrium between bone formation and bone destruction does accordingly not occur In the early stages of osteitis fibrosa, giant cells are always present in large numbers, that is to say that, in the view of Geschickter and Copeland, solitary bone cysts may in many cases be regarded as healed giant-cell tumours The healing occurs in the metaphysis in which the cortex and periosteum are thick, but not in the epiphysis where the cortex and periosteum are thin, accordingly, giant-cell tumours involve the epiphysis and cysts the metaphysis Giant cell tumours occur only in bones preformed in cartilage, but the maxilla and mandible form exceptions from this rule That giant cell tumours do occur in these bones is explained by Geschickter and Copeland as follows

The shedding of the deciduous teeth is brought about by proliferation of giant cells arising from the periosteum and resorbing the roots of these teeth Epulis occurs in relation to this resorption of the deciduous teeth, according to Geschickter and Copeland this explains why epulis is most frequent in children and adolescents, never develops before secondary dentition, and never involves the posterior molars

In contrast to these authors Jaffe, Lichtenstein and Portis (1940) claim that a genuine giant-cell tumour exists which must be regarded as a neoplasm Based on its histological structure, mainly on differences in stromal cell detail, they classified this tumour into three grades

Grade I has spindle-shaped stromal cells and few or no mitoses

Grade II constitutes a somewhat varied group, ranging from tumours in which the stromal cells show only slight atypism to those in which they are strikingly atypical

Grade III has a large number of closely compacted anaplastic stromal cells presenting an irregular whorled arrangement, and according to the authors, tumours of grade III tend to recur and metastasize Sharp, Bullock and Hazlet (1956) report that others have found little correlation between the clinical behaviour and the microscopic appearance

In addition, some other giant-cell tumour variants exist, for example, epulis, on which Jaffe *et al.* comment as follows:

"The problem of explaining the evolution of giant-cell epulides is a rather difficult one, involving, as already suggested, the old bedevilling controversy of neoplasia versus posthæmorrhagic reparative granuloma. The difficulty may be resolved in part if one regards the epulides as probably constituting a mixed group of lesions with a varied genesis, which is reflected in their microscopic appearance."

On the relation between the epulides and the giant-cell tumour Jaffe *et al.* write:

"The epulides have in common a rather vascular connective tissue ~~and~~ an appreciable number of multinuclear giant cells.

There are also certain epulides with a more richly cellular stroma composed of ovoid and spindle-shaped cells, and there are numerous giant cells whose nuclei resemble those of the stroma and may even present an appreciable number of mitoses. Such an epulis may develop as an outgrowth of a central giant-cell tumour of the jaw, but may also arise as an independent growth representing the periosteal counterpart of the giant-cell tumour of bone."

Bernier and Cahn (1954) reported 43 cases of epulis. The histological picture was found to differ from that of genuine giant-cell tumour; there were fewer giant cells, which were not scattered diffusely but were arranged in irregular clusters. The stroma was characterized by fibrocytes arranged in fibrous areas, and the entire lesion was surrounded by a zone of connective tissue. The presence of blood pigment was interpreted as indicating a previous hæmorrhage, which was taken as evidence in support of the assumption that trauma plays a part in the pathogenesis. The authors therefore used the designation "peripheral reparative giant-cell granuloma" instead of tumour. In more than one half of the cases a history of trauma was revealed, most frequently in the form of dental extraction.

Sharp *et al.* (1956) classified the giant-cell lesions of the jaws as follows:

1. Central giant-cell tumour.
2. Central giant-cell reparative granuloma.
3. Peripheral giant-cell reparative granuloma.

The distinction between tumour and granuloma is based on the differences in the microscopic appearance as described by Jaffe *et al.*

The distinction between central and peripheral lesions is based both on the clinical and radiographic appearance.

The peripheral lesion is assumed to arise from the periosteum, from which it grows towards the gingiva, so that it becomes visible before radiographic bone changes develop, whereas the central form arises in the osseous tissue, growing expansively and producing a rounded radiolucent zone in the jaw (soap-bubble appearance)—usually unilocular, only in rare instances with the lamina dura preserved everywhere However, radiographic changes which are specific for central giant-cell tumours of the jaws do not exist (Sonesson, 1950, Lichtenstein, 1952, Waldron, 1953).

Personal Investigations

In the Radium Centre and the Department of Otolaryngology of Aarhus Kommunehospital a total of 150 cases of tumour of the gingiva were treated within the years 1935-1957 Among these, histological examination revealed *giant cells in 26 cases, which are therefore here referred to as giant-cell epulis.* The patients were 15 females and 11 males, whose ages ranged from 7 to 78 years, the average ages for females and males being 38 and 34 years, respectively, 14 of the patients were over 30 and only four under 20 years of age, which speaks against the assumption of Geschickter and Copeland that secondary dentition should be of importance in the development of epulis

Localization

Of the epulides, 17 involved the upper jaw, including 12 on the right side, and 9 the lower jaw This distribution is not in agreement with other reports, in which it is usually stated that the upper and lower jaws are equally affected

In not less than five cases did the epulis involve the molar region, *which is also in disagreement with the theories of Geschickter and* Copeland

Trauma

In 19 of the patients a history of previous trauma* could not be elicited In five cases, the epulis developed at a site where a tooth had been extracted shortly (1-12 months) before the lesion was noticed, and the patients attributed the development of the tumour to the extraction In two patients, a trauma was followed by increased growth of the tumour. About one half of the patients wore dentures

Symptoms

Epulis was usually completely asymptomatic In most cases, the tumour was discovered quite accidentally, and its growth had occurred without producing any symptoms Only one patient complained of tenderness, and two had experienced oral bleeding In several cases, it was the dentist who had first noticed the tumour

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* As distinct from Bernier and Cahn who revealed traumata in 53 per cent of their cases we did not include peridental treatment irritation from prostheses injudicious use of toothpicks etc , under the concept of trauma

Pathology

At the physical examination, the tumour presented a highly variable appearance. Its diameter varied from 0·5 to about 5 cm. The colour was usually bluish red. The tumour was described as an elastic, irregular mass of a fleshy consistency, and usually broad-based (Fig. 1). In two cases, expulsion or loosening of teeth had occurred in the tumour region. Occasionally, ulceration of the tumour surface was present.

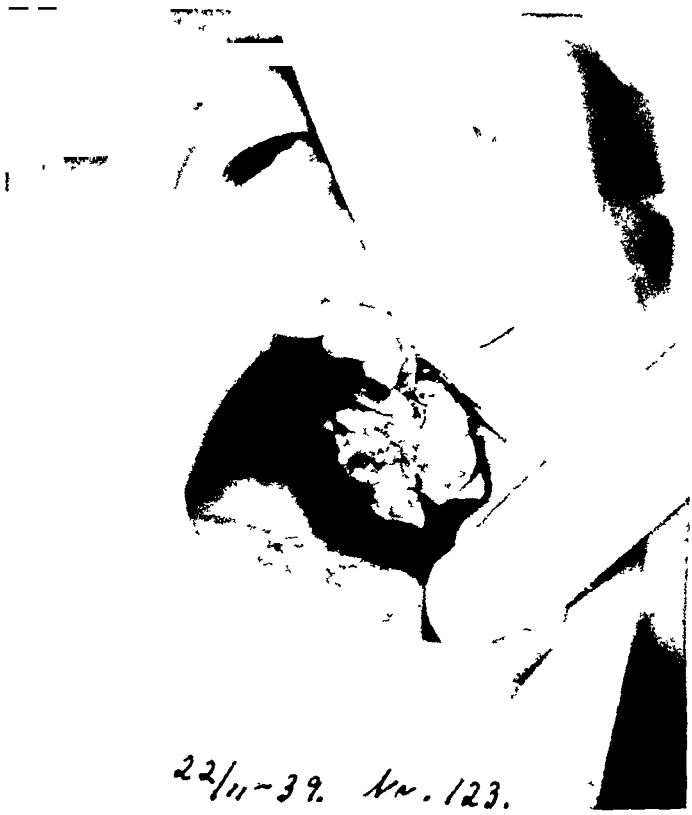

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FIG. 1

Large giant-cell epulis of the left upper jaw

Radiographic Appearance

Radiographic studies were performed in 16 of the 26 patients. No abnormalities were revealed in eight, while the remaining eight showed a radiolucent zone of varying size in the alveolar process of the involved area. This was also observed in several patients with very small and apparently quite superficial tumours. Thus, none of the cases revealed the *aforementioned changes which (according to Sonesson) are characteristic, although not pathognomonic of central giant-cell tumours.*

It must be assumed that it is not always possible to distinguish between peripheral and central tumours.

Histology

The histological examination with a view to a possible differentiation into several groups of pathohistological pictures was performed by Dr. Tage Lund, of the Institute of Pathological Anatomy, Aarhus Kommunehospital.

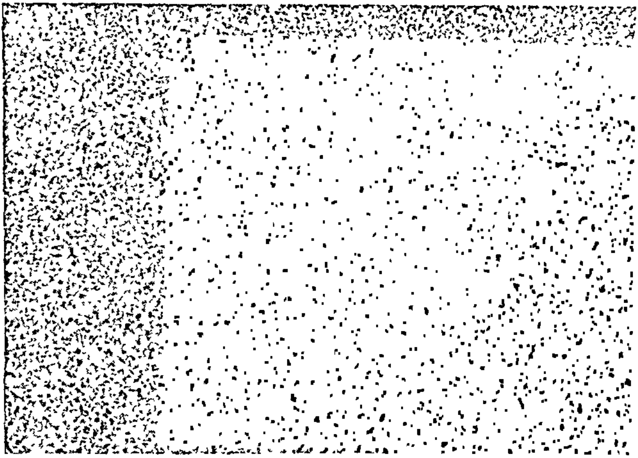

FIG 2

Section from giant-cell epulis The giant cells are scattered diffusely in a uniform stroma ×48

This examination revealed a highly uniform picture in all the tumours except three, which differed from the main group.

The typical histological picture showed a stroma composed of mononuclear spindle-shaped or ovoid cells with rounded or oval nuclei. The cells were closely compacted with a sparse intercellular substance. No fibrous areas were seen. Numerous multinuclear giant cells were scattered diffusely in the stroma. The nuclei of the giant cells were most frequently collected towards the middle of the cells; they were rounded or oval, resembling those of the stroma (Figs. 2 and 3).

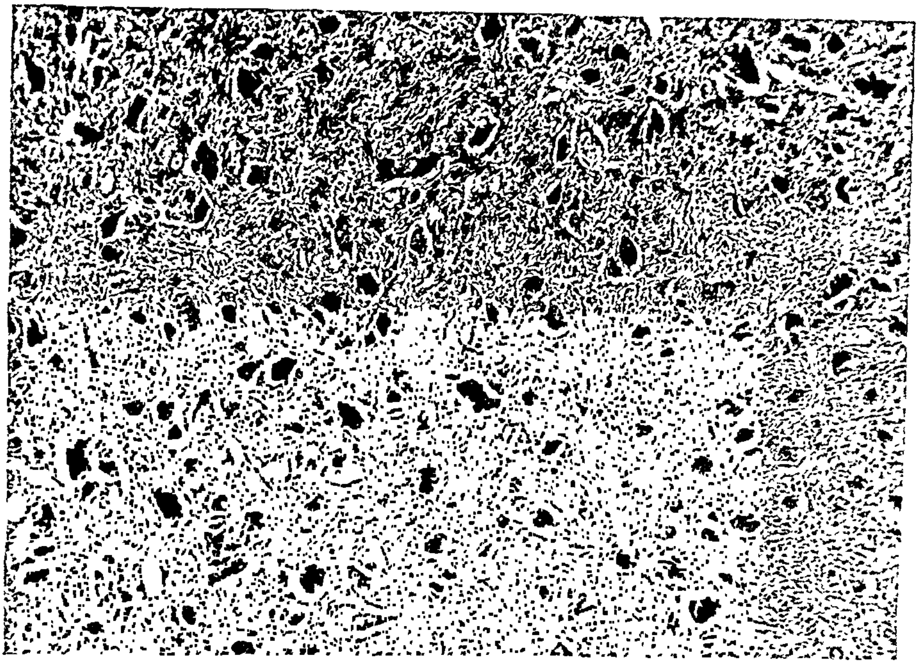

FIG. 3.
Section from giant-cell epulis. The stromal cells are closely compacted; only sparse amounts of intercellular substance are present. × 192.

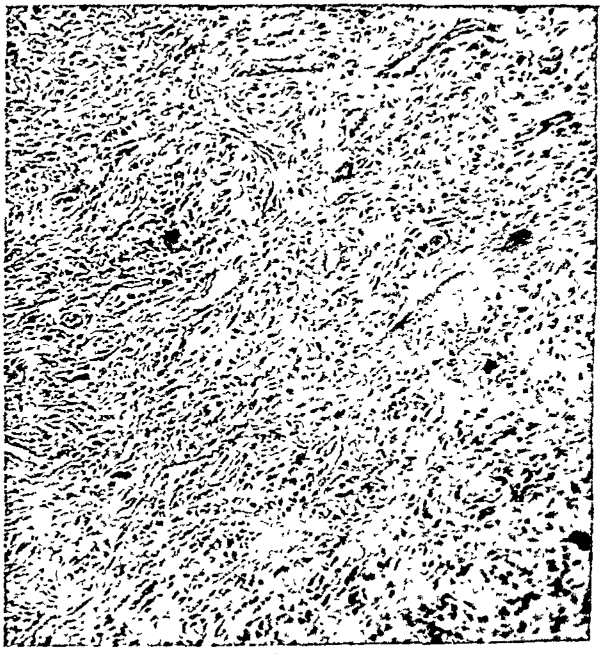

FIG. 4.
Section from a fibrous epulis, showing few scattered giant cells. × 192.

The tumour tissue showed abundant vascularization, and hæmorrhage was frequently seen. A few tumours revealed bone tissue.

Two tumours showed numerous fibrous areas in the stroma. The giant cells were here collected in irregular clusters, thus displaying the same histological picture as described by Bernier and Cahn. One tumour was composed almost exclusively of fibrous tissue; only towards the periosteum a few giant cells were seen (Fig. 4).

Thus, in contrast to Bernier and Cahn, we found in all cases except three the typical picture of a genuine giant-cell tumour just as is seen in the long bones and similar to grade I described by Jaffe *et al.*

Treatment

Small epulides without bone involvement may be removed locally. In the presence of bone involvement, the removal must be combined with dental extraction and curettage or electrocoagulation. Large central tumours require excision of the bone tissue with complete exposure of the area involved. Thoma and others claim that surgical treatment is adequate, and that recurrence may be due to inadequate removal.

Jaffe *et al.* maintained that in the treatment of giant-cell tumours there seems to be some evidence that in certain cases irradiation as an adjuvant to curettage may even stimulate recurrence or increase the aggressiveness of the lesion.

In the present series, seven patients were given surgical treatment alone consisting in extirpation of the growth. In the remaining patients, surgical treatment was combined with postoperative irradiation by radium needles inserted into the cavity. Three patients were treated with radium irradiation only, and one was given pre-operative roentgen irradiation.

The lesion did not recur in any of the seven patients who had received surgical treatment alone. Recurrence was experienced in one patient of the group given surgical treatment plus irradiation and in one of the three patients who were treated with radium irradiation alone. In both cases, the recurrence disappeared after renewed treatment, and all the patients have since been free from recurrence, in most cases for more than 3 years. The recurrent lesions showed the same histological picture as the main group of the original tumours.

The surgical measures applied were simple extirpation, sometimes followed by curettage of the tumour-bearing area or electrocoagulation. In two patients, extraction of teeth were necessary. The result of this purely surgical treatment seems to be fully satisfactory. One of the cases treated with radium gave rise to osseous necrosis with sequestration. It may be said that supplementary radium irradiation does not improve the result.

Case Report

It may be of interest to give a brief description of one of the case histories because it illustrates the part played by trauma in the development of epulis.

The patient was a 9-year-old boy, who had previously been in good health, without any symptoms from the mouth or teeth. One day, he fell, hitting the right side of the mouth against an iron bar. Some bleeding occurred, and the right canine tooth of the upper jaw loosened. Two days later swelling of the right cheek developed, it continued to increase in size, for which reason the patient was admitted to hospital three weeks later. Clinical examination revealed a bluish red tumour of a fleshy consistency, about 4 cm. in diameter, in the right upper jaw, strongly suggestive of malignancy (Fig. 5). Radiographic examination revealed a large, expansive, space-occupying lesion of the right superior alveolar process with displacement of the developing dental tissue and elimination of the normal osseous border in the right side of the gingiva (Fig. 6). The levels of serum calcium and serum phosphorus were normal.

Fig. 5

Boy, aged 9 years, with a large giant-cell epulis of the right upper jaw

A biopsy specimen showed typical genuine giant-cell tumour with numerous scattered giant cells embedded in a uniform stroma without fibrous areas, but with distinct traces of hæmorrhage. In view of the close relationship in time between the trauma and the unusually rapid growth of the tumour, it was tempting to assume that the trauma was the causative factor However a renewed examination of some radiographs taken three days after the trauma showed a small cyst-like radiolucent area around the lateral incisor on the right side As such a cyst could not possibly have developed within the space of three days, this observation shows that the tumour must have been present before the trauma was sustained but its growth has probably been accelerated by the trauma The lesion was a giant cell tumour presumably of central origin

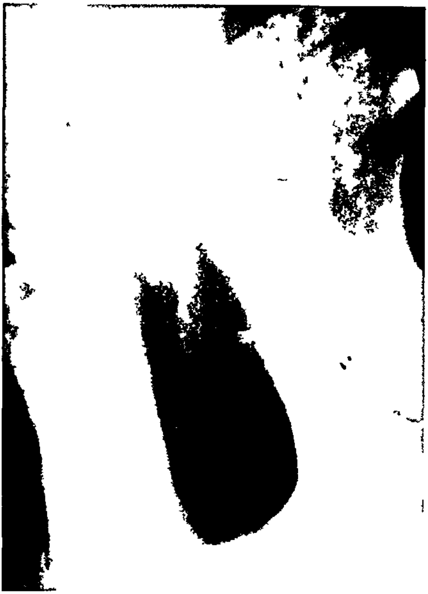

FIG 6
Radiograph of the upper right alveolar process showing an area of osseous destruction around the lateral incisor

Discussion

Clinically, the appearance of giant-cell epulis shows very wide variations It is difficult, with certainty, to distinguish between central and peripheral epulides, since there are no radiographic changes which can be said to be specific for central giant-cell tumours, and since even small superficial epulides may give rise to radiolucent areas in the bone.

Epulis has *a favourable prognosis.* In the present series, only two recurrences were encountered after treatment. Surgical treatment gave good results, which were not further improved by postoperative supplementary radium irradiation.

The distribution and localization of the lesions in the present series do not reveal any evidence in support of the assumption advanced by Geschickter and Copeland that *the osteoclastic activity in the resorption of the deciduous teeth* should play a part in the development of epulides.

Only in a minority of patients can epulis be related to a *trauma,* and even in such instances the importance of traumatic injury must be regarded as problematic, since it has been shown in one case that the tumour was present before the trauma was sustained. *Thus, it is unlikely that epulis is generally due to traumatic injury,* but it will often be so that a trauma calls attention to the tumour and may accelerate its growth because of bleeding and disturbances in the blood supply.

In most cases of the present series, *epulis could not be distinguished histologically from other genuine giant-cell tumours.* Like the latter growths, epulides show a uniform picture with a fairly dense stroma composed of irregularly arranged, spindle-shaped or ovoid cells. Fibrous areas are absent. Giant cells are present in large numbers and are scattered diffusely in the stroma. It is therefore likely that most cases of epulis, like giant-cell tumours of the long bones, are genuine tumours.

The present series revealed only three cases with a different histological composition with many fibrocytes arranged in areas embedding giant cells in irregular clusters. This picture is more similar to that described as "peripheral reparative granuloma".

Thus, there are various forms of giant-cell epulis differing in histological composition and, probably, in genesis.

Considering the histological composition which cannot be distinguished from that seen in giant-cell tumours of the long bones and the fact that the series did not reveal any definite evidence in support of the ætiological importance of traumatic injury but, on the contrary, in one case showed that the tumour was present before a trauma called attention to it and gave rise to increased rapidity of growth, *most giant-cell epulides must be interpreted as genuine tumours.*

Summary

An analysis of 26 cases of giant-cell epulis is presented.

The importance of trauma in the development of epulis is discussed. In one case in which the tumour was diagnosed shortly after a severe trauma, it could be demonstrated that the tumour had been present before

the trauma was sustained, for which reason it was assumed that the trauma had merely accelerated the growth of the tumour.

Radiographically, osseous changes can be demonstrated even in small epulides.

Histologically, most cases of epulis cannot be distinguished from genuine giant-cell tumours.

The analysis shows that several forms of giant-cell epulis exist, possibly differing in genesis. The majority of the cases in the present series must be interpreted as genuine giant-cell tumours.

Surgical treatment gives good results.

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MAJOR SURGERY OF THE LARYNX UNDER INDUCED HYPOTHERMIA

A PRELIMINARY REPORT

By R. W. BAILIE, A. K. BOYLE, and A. MACIEJEWSKI (Glasgow)

THERE have been many reports in recent years on the use of radical surgery for pharyngo-laryngeal carcinoma. In these the necessity of combining unilateral or bilateral block dissection of the neck, preferably by the monoblock technique, with the excision of the cancer has been emphasized. Among others, Wookey (1948), Owen (1954), Raven (1954 and 1957), Ormerod and Shaw (1956), Miles Foxen (1957) and Hayes Martin (1957), have described their experiences with extensive surgical procedures for malignant growth in the region of the larynx.

Radical ablative surgery requires considerable time for its execution. The patients are often poor operative risks. The method of anæsthesia to be adopted is a consideration of the first importance.

Local analgesia may be used. It is not, however, the method of choice in this country. General anæsthesia is preferred.

The use of induced general hypotension for total laryngectomies has been the subject of several publications.

We have employed induced general hypothermia in five cases.

General Considerations

The patients requiring operation were from middle and older age groups.

The tumour originated in the lower pharynx in three cases and from the epiglottis in one. In the fifth case, the growth represented the recurrence, after cobalt-beam therapy, of an intrinsic laryngeal carcinoma.

There was spread to the regional lymph nodes in four instances, suggested pre-operatively in three by palpable cervical glands and discovered at operation in the other.

Dysphagia and respiratory obstruction were present, in varying degree, in every case. Recent weight loss was a constant finding.

Additional disease existed in the form of chronic bronchitis and emphysema, arterial hypertension or anæmia. In two of the patients all three conditions were present.

The surgery proposed on three occasions was pharyngolaryngeal resection with block dissection as required and one-stage reconstruction where possible

Of the difficulties expected during and after operation, two demanded the closest attention The first was the maintenance of a perfect airway throughout, and the second, the control of blood loss

The respiratory obstruction occasioned by the growth itself was certain to be increased during the surgical manipulations Furthermore, it was essential to prevent blood entering into the trachea and bronchi and to provide for the aspiration of tracheobronchial secretions Delivery of the anæsthetic gases through a cuffed and unkinkable tube was indicated Elective tracheostomy would have simplified the conduct of general anæsthesia but would have interfered with the siting of the final tracheostomy according to the local conditions obtaining Transoral intubation and final tracheostomy was decided upon It was agreed that, on those occasions when the size and position of the tumour or its vascularity was such, that intubation after induction of anæsthesia would be hazardous, laryngotomy or tracheotomy would be carried out under local analgesia, as a preliminary to general anæsthesia

It was anticipated that loss of blood would be heavy, constituting a source of danger to the patient and an impediment to the surgeon Purposely lowering the blood pressure by autonomic ganglion blockade to secure an ischæmic field of operation was considered and rejected We have never been entirely convinced of the safety of induced hypotension, wherein the blood flow and, therefore, oxygen supply to the tissues is reduced without the protection of a diminished oxygen demand by the tissues In induced hypothermia the oxygen needs of the tissue cells are lowered and the margin of safety for cell vitality is temporarily extended in the presence of anoxia Our previous experiences with light-plane hypothermia (body temperature range 35° to 30°C) in unfit subjects undergoing resections of the pelvic viscera attended by the same danger and inconvenience from hæmorrhage, has been most encouraging and prompted its adoption for these cases

Post operative vomiting which might well lead to disruption and contamination of recently sutured wounds had to be prevented if possible The anti-emetic properties of the anti-histamine drugs used in our hypothermic technique are well recognized

Pre-Operative Assessment and Preparation

In addition to a complete physical examination by a consultant physician and *roentgenological examination of the chest, the following* investigations were made routinely complete blood count, hæmoglobin estimation, plasma electrolytes and blood urea estimation. Respiratory function tests and electrocardiographic recordings were made.

The locality and extent of the tumour and the degree of impairment of the airway were discussed with the surgeon and a mutually satisfactory anæsthetic management determined.

The general nutrition of the patients had to be improved before operation and an enriched diet, easy to swallow and assimilate, was prescribed. Even with this semifluid diet, the greatest and most persistent efforts at deglutition were required of two of the patients. Payling Wright (1954) has pointed out that repeated attempts at swallowing against obstruction may act in a "massaging manner" on the tumour, resulting in stimulation and local dissemination. General anæsthesia would have been necessary in these two cases to pass a feeding tube into the stomach, since attempts to do so while they were conscious were not successful.

As a prophylaxis against post-operative pulmonary complications, breathing and coughing exercises were taught by the physiotherapist, well in advance.

Low hæmoglobin values were corrected by blood transfusion, sometimes of packed red cells, on the last pre-operative day.

A good night's rest was ensured with butobarbitone 195 mg., followed two or three hours later with chloral hydrate 1 g. by mouth, or paraldehyde 5 ml. by intramuscular injection, prior to operation the following morning.

Anæsthetic Technique

The technique of induced hypothermia with a few minor variations was the same in all cases.

Premedication, consisting of promethazine hydrochloride 50 mg. by intramuscular injection or promethazine-8-chlortheophyllinate 50 mg. by mouth two and a half hours before cooling, and pethidine hydrochloride 100 mg. with hyoscine hydrobromide 0·43 mg. intramuscularly one hour later, was given.

An intravenous drip infusion of cooled normal saline was then set up as a vehicle for the administration of the so-called "lytic cocktail", comprising chlorpromazine hydrochloride 50 mg., promethazine hydrochloride 50 mg., and pethidine hydrochloride 100 mg. made up to 10 ml. of solution with normal saline. This was injected in divided doses over a period of thirty minutes.

Anæsthesia was induced with thiopentone sodium, 100 to 300 mg. Tracheal intubation transorally under direct vision was performed with the aid of succinylcholine chloride, 50 to 80 mg. Maintenance was with nitrous oxide and oxygen (5l./3l.) with manually controlled respiration. To facilitate the control of respiration and to eliminate shivering d-tubocurarine chloride was administered fractionally.

Cooling, conducted in a screened corner of the ward, was by the application of icebags to the uncovered patient, aided by cold draughts from a widely-opened window. Cautious turning of the patient a few degrees from side to side, periodically, prevented pooling of the blood in dependent parts.

Recordings of pulse rate, blood pressure, rectal temperature and ambient temperature were made at 10 minute intervals from the start and throughout the surgery, together with regular electrocardiographic tracings.

When the body temperature had fallen to 33° to 32°C., the surface cooling was suspended. An after-fall of body temperature of 2° to 3°C. always occurred.

The orotracheal tube remained in situ until that stage of the operation had been reached when the surgeon required its removal. Withdrawal was synchronized with the insertion of a short cuffed tube through the larynx, already opened or through a tracheal incision, previously made. The short tube of Sjöberg or Radcliffe type, together with its suction union, endotracheal catheter mount and rubber connection, sterilized as a complete assembly, had been placed on the instrument trolley for use by the surgeon when required.

Blood replacement was adjusted to blood loss. Suction was not employed, all blood shed being collected on dry swabs and weighed. To the calculation of blood loss on a weight/volume coefficient a correction factor of +10 to 20 per cent. was applied. Table I. gives the actual volumes of whole blood transfused.

TABLE I

DURATION-TIME OF OPERATION AND TOTAL VOLUME OF WHOLE BLOOD TRANSFUSION

Case No	Duration of Operation (mins)	Total Volume of Blood Transfused (mls)
1	280	1,620
2	185	1,350
3	310	1,620
4	245	1,080
5	225	1,350

Particular care was exercised with hæmostasis throughout the operative field before reconstruction was begun.

Tracheobronchial toilet was regularly performed.

Post-Operative Management

At the conclusion of the operation it was verified that:

(1) Blood loss was fully restored.

(2) The blood pressure was stable at a safe level (100 mm. Hg systolic, or above).

(3) The airway was unobstructed.

(4) The neuromuscular block was completely reversed.

(5) Central depression of respiration had been abolished.

Assistance was provided until all doubt as to the efficiency of natural respiration had been dispelled.

Rewarming was allowed to take place spontaneously. On return to bed the patient was covered by a single sheet and the appearance of shivering was the indication for the application of additional covering.

The charting of pulse rate, respiration rate, blood pressure and rectal temperature was continued every 10 minutes for the first 12 hours, and thereafter, less frequently.

All patients were placed in the Fowler position as soon as it was deemed safe and all had wide-spectrum antibiotic cover from the outset.

Removal of tracheobronchial secretions by suction, at least every half-hour was instituted from the beginning. After the first 24 to 48 hours, two or three millilitres of normal saline were instilled into the trachea immediately before every third or fourth aspiration in an endeavour to keep the secretions loose and to stimulate coughing. Atropine was avoided throughout.

The breathing and coughing exercises were resumed as soon as feasible.

Analgesics and sedatives, because of their depressant effects on the cough reflex and on the respiratory centre, were to be exhibited in the early recovery period only when essential. Two of the patients were restless and complained of pain. Both were given paraldehyde 5 ml. intramuscularly. In addition, the first had pethidine hydrochloride twice in 50 mg. doses subcutaneously, and the second pethidine hydrochloride 50 mg. and levallorphan tartrate 0·625 mg., once repeated.

The fluid intake and output were measured and balanced.

Early ambulation was encouraged.

Difficulties encountered during Anæsthesia and Hypothermia

During the induction and maintenance of anæsthesia and hypothermia the problems met with were as follows:

1. Difficulty in visualizing the glottis.

In three of the cases the larynx was grossly distorted and displaced by the growth and identification of the glottic opening was difficult. It might well be impossible to intubate perorally.

2. Lability of the Circulation.

The sudden and marked variations in blood pressure which occurred in two of the cases, during cooling and during operation, were ascribed to abrupt changes in posture, carbon dioxide retention and surgical interference with the sino-aortic nerves

3 Lack of control of body temperature

This did not cause any great concern It was not proposed to lower the patient's temperature below 30°C and allowance was made for "after-fall" by discontinuing the cooling process at 33° to 32°C, as already stated.

Electrocardiographic Changes

The electrocardiographic, clinical and radiological examination of the cardiovascular systems of these patients showed no abnormality before operation

During the entire period of induction and maintenance of hypothermia electrocardiographic monitoring was carried out with a Mingograph (Elinquist) direct-writing electrocardiograph Lead 2 was used routinely

In these five cases no arrhythmia was noted There was a slight fall in heart rate in all and in two there were increased P R intervals An increase in the corrected QT rate was a constant In every record changes developed in the early part of the ST segment, which on three occasions consisted merely of a widening of the base of the QRS complex whereas in two other records it amounted to the development of a separate upward deflection early in the ST segment, clearly separated from the QRS complex In the cases with this additional wave the body temperatures, when it was recorded, were as high as 30° and 33 6°C respectively When only QRS widening was noted it was seen when the temperatures were 35 8° and 32 2°C These changes are well recognized but usually occur consistently only on cooling to 31°C or lower The changes have been the subject of a recent special study by Emslie-Smith (1956)

Post-Operative Complications

There have been no deaths

The non-fatal complications, related to anæsthesia and hypothermia, that have occurred following operation are set out in Table II

TABLE II

Post Operative Complications

Type	No of Cases
Restlessness	2
Respiratory Obstruction	2
Pyrexia	2
Surgical Emphysema	3

The respiratory obstruction which developed in the first case of the series on the third day was attributable to two factors:

Obtundation of the cough reflex by pethidine and paraldehyde, administered for pain and restlessness.

Inefficient aspiration of secretions from the lower airway, the suction catheter not being inserted sufficiently deeply.

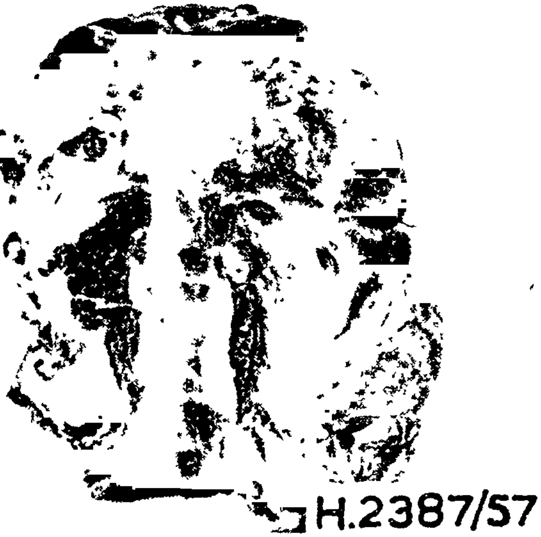

FIG. 1.
Specimen removed at operation in Case 3, showing the large tumour mass lying above and to the left of the glottic opening. The epiglottis has been almost wholly destroyed

Post-hypothermic rise of temperature never exceeded 38·2°C. It responded readily to tepid sponging.

The surgical emphysema observed in three of the cases was confined to the neck or to the neck and upper pectoral region. Clearance was rapid.

Case Reports

Case 1. A male patient, aged 48 years, was first seen in April, 1957. He had a history of huskiness and cough for one year and of "food tending to stick in his throat" over the last two months.

On examination an ulcerated mass was present on the epiglottis, obscuring the vocal cords. The latter were visualized only on direct laryngoscopy and, even then, a clear view was not obtained.

Biopsy revealed the mass to be of the nature of a squamous carcinoma.

Total laryngectomy, with removal of the hyoid bone and the pre-epiglottic tissues and left radial block dissection of lymph glands, was performed by mono-block technique. The enlarged cervical glands on the left side, palpable pre-operatively, showed no evidence of malignant invasion on microscopy.

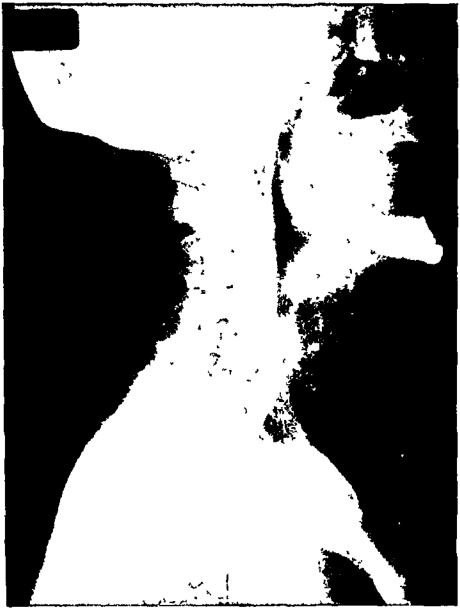

FIG 2

Lateral radiograph taken 18 days before operation in Case 3, demonstrating a spherical tumour mass, centred at hyoid bone level

His condition is very satisfactory with no evidence of recurrence to date (February, 1958).

Case 2. A man, aged 35 years, was admitted to hospital in September, 1957, with the complaint of burning and discomfort in his throat for four months, of loss of weight (10 Kg.) and, in the last four or five weeks, of dysphagia.

Examination showed an ulcerating tumour involving the whole lumen of the post-cricoid area and biopsy defined it as a squamous carcinoma. The left vocal cord was immobile and an enlarged lymph gland could be felt in the left hyoid region.

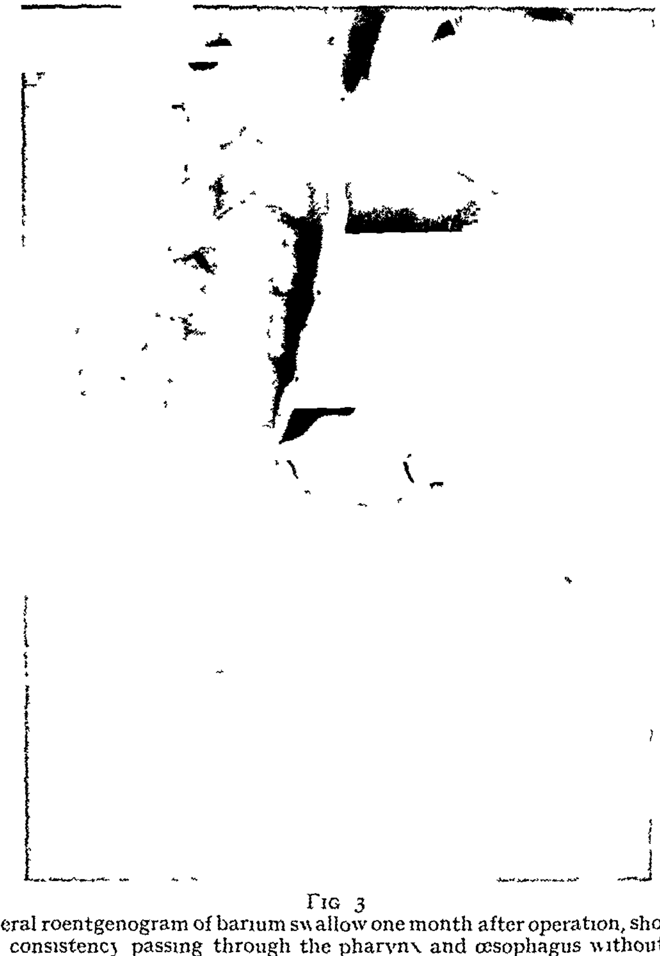

FIG 3

Case 3 Lateral roentgenogram of barium swallow one month after operation, showing barium of medium consistency passing through the pharynx and œsophagus without evidence of obstruction

First-stage pharyngo-œsophago-laryngectomy was carried out with left radical block dissection. The first 2·5 cm. of the œsophagus were removed. The glands just below the hyoid bone were very adherent and their excision very difficult. They proved to be heavily infiltrated with cancer cells.

His convalescence was uninterrupted but there is now evidence of recurrence in the region of the involved glands.

Case 3. A 59-year-old woman, who had been under treatment for hypochromic anæmia with histamine-fast achlorhydria for six years, was referred to the E N T surgeon (R W B) as complaining of discomfort in her throat and slight dysphagia for the past nine months, having lost over 12 5 Kg during that period A growth, extending from just below the lower pole of the right tonsil to the lower part of the pyriform fossa, was found which had apparently caused wide destruction of the epiglottis

It proved to be a squamous cell carcinoma

Pharyngo-laryngectomy, including resection of a large part of the base of the tongue, extensive right-sided block dissection of glands and removal of right lobe of thyroid gland, was effected by monoblock technique A strip of mucosa some 2 cm in width, on the left postero-lateral pharyngeal wall, was preserved and its edges approximated over a per-nasal indwelling stomach tube to reconstruct the pharynx and the U-shaped skin flap replaced and sutured over it

Recovery was uneventful Despite considerable glossal paresis, swallowing, except for some solids, is now quite free There has been no recurrence

Case 4 A woman, aged 68 years, was admitted in December, 1957, complaining of dysphagia for solid food for six months and of "wasting" Her body weight was 30 5 Kg She had suffered from dyspepsia for twenty years

A mass was present in the right pyriform fossa, extending down into the post-cricoid region and below

The pathological report upon the biopsy specimen was that it was a squamous carcinoma

First-stage pharyngo œsophago laryngectomy was performed and it was found necessary to remove 6 cm of the œsophagus and the whole of the thyroid gland, the left lobe of which had been invaded Right radical block dissection was included in the one block of tissues removed Left block dissection was deferred, frozen-sections of two small lymph glands from the carotid sheath displaying no malignancy Fixation of the Gluck tube inserted in this case proved too much of a problem and it was discarded

Stomach-tube feeding has been hindered by regurgitation, attributable to cardiospasm and surgical treatment of the latter is under consideration

Case 5 A male patient, aged 45 years, had been treated in September, 1956, by Cobalt-beam therapy for a well differentiated squamous carcinoma involving most of the right vocal cord Following a good initial response to the therapy, considerable swelling of the right half of the larynx developed in the later months of 1957 There was no response to antibiotic treatment Weight loss over the last year was some 15 Kg

Biopsy from the larynx showed malignant infiltration

Total laryngectomy was done in January, 1958, using a wide collar incision because of the previous irradiation. No block dissection was performed as no glands could be palpated.

Although suturing was meticulous during closure of the pharynx, a pharyngeal fistula developed a few days after operation. His general condition, however, has been good throughout and the fistula now appears to be closing spontaneously in the fourth post-operative week.

Discussion

When malnutrition is gross and dysphagia pronounced a polyvinyl tube of adequate diameter, passed through the nose or mouth into the stomach at the time of the laryngoscopy (and/or œsophagoscopy) under general anæsthesia for evaluation and biopsy, will resolve the problem of pre-operative feeding. Should this prove impracticable, gastrostomy or jejunostomy should not be delayed.

Too much reliance must not be placed on dexterity at intubation after thiopentone and scoline in patients with respiratory obstruction. When the airway is seriously impaired the need for preliminary tracheostomy or pre-operative laryngotomy or tracheotomy must not be disregarded.

It is possible that the introduction of a tube into the trachea, traversing the neoplasm en route, may result in implantation of tumour cells in the trachea, but, unless patency of the airway at all times is assured, hypoxic episodes are unavoidable. In the absence of a preliminary tracheostomy and without an intratracheal tube, the conditions under general anæsthesia are such that an emergency tracheotomy for the urgent relief of respiratory obstruction may be required at an early stage. This is not an ideal start for an operation of great magnitude in a seriously ill patient. Moreover, recurrence around the final tracheostome or beyond is much more likely to be due to division of the trachea above the lowest level of submucous extension of the growth or to spread along the paratracheal lymphatics into the tracheo-œsophageal groove (Ormerod and Shaw, 1956).

We have had to deal with very unfit patients subjected to the most radical ablative surgery, lasting up to five hours. Under such circumstances even the most diligent support of the cardiovascular system by intravenous transfusion will not always prevent the development of surgical shock when conventional techniques of general anæsthesia are employed. Nor will it deal with the obscuring effect of hæmorrhage.

The risk from myocardial or cerebral ischæmia during hypotensive anæsthesia increases with the duration time. In prolonged operations, under induced hypotension, particularly in patients with a vulnerable cardiovascular system, death from cardiac arrest or necrotic lesions of the brain have been reported with alarming frequency.

In this small series of cases conducted under induced hypothermia the operative site may not have been entirely "dry" but the volume of blood lost, as measured, has been small, visibility has been unimpaired and surgical progress unimpeded The operation has never had to be modified in extent nor completed in urgency because of deterioration in the patient's condition Recovery from anæsthesia and restoration of protective reflexes have been rapid and there has been no post-operative vomiting Freedom from pain after operation and ready participation of the patients in their early post-operative management has been a striking feature in four of the cases In the remaining one, pain exerted a distracting influence and this patient was restless and unco-operative in the early recovery phase Pethidine hydrochloride was the drug selected to combat pain Its respiratory depressant effect may be antagonized, without interference with its analgesic effect, by levallorphan tartrate (Hewer and Lee, 1957)

Rewarming presented no problem

Summary

The use of light-plane hypothermia in five cases undergoing major surgery for malignant growth involving the larynx is recorded

The pre-operative preparation, surgical and anæsthetic techniques and post-operative management are described

The problems encountered are considered

We have been impressed by the excellent condition of the patients, during and after, the extensive and lengthy operations performed

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CLINICO PATHOLOGICAL INVESTIGATIONS ON ALLERGIC RHINITIS

By A Y AWNY E D EL SAMMA and A A SOLIMAN (Cairo)*

IN a previous study on patients suffering from bronchial asthma (Awny *et al* 1957) certain laboratory findings were encountered which were of valuable interest to both the clinician and the laboratory worker

These findings initiated a stimulus to conduct the same type of research on a group of patients suffering from a similar allergic state namely allergic rhinitis This is an allergic condition involving the nasal mucous membrane characterized by paroxysmal rhinorrhœa, sneezing, nasal obstruction and itching of the nose

The underlying cause of the condition is a state of hypersensitivity of the nasal mucosa Allergens are commonly inhalants as dust, animal dander, etc Food, physical agents and bacteria are less frequent causes Neurogenic, psychogenic, endocrine and metabolic disturbances are contributory factors The nasal mucous membrane is typically pale bluish and œdematous The discharge is thin and watery Marked œdema of the nasal mucosa constitutes the pathology in the early stages of the condition The bogginess and œdema of the mucous membrane lead to the formation of polypi which may fill the nose and some of the sinuses The mouth of the subepithelial glands become occluded and they undergo cystic degeneration The diagnosis of the condition depends on the history and physical findings and it has to be differentiated from similar conditions like the common cold and sinusitis

The object of this research is an attempt to find out further laboratory criteria which would help in making differentiation based on a sounder basis Together with this a clinical study of the results of various lines of treatment for the allergic cases was included

The following investigations were carried out on a group of 37 patients in which 25 cases are clinically diagnosed allergic rhinitis and in 12 cases infection was the predominant etiological factor

(1) Examination of smears from the nasal mucous membrane and from

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nasal discharge. The smears were stained by methylene blue, Gram stain and Leishman stain.

(2) Examination of the total and differential leucocytic counts of the blood.

(3) Determination of erythrocyte sedimentation rate by the Westergren method.

(4) Examination of the stools and urine for parasitic infections.

(5) Determination of the urinary excretion of 17 Ketosteroids by the method adopted by Robbie and Gibson (1943) and Gallow *et al.* (1938).

(6) Determination of the serum protein fractions by the photoelectric calometric methods using the biuret technique and salting out procedure.

(7) Estimation of the serum sodium and potassium in m. eq. per litre by the flame photometer.

The idea of selecting these tests was to investigate the degree of disturbance (if present) in:

(*a*) The endocrine function particularly of the suprarenal glands.

(*b*) The electrolyte balance and possible rôle of the disturbance in the pathogenesis of the hypersensitive state of the nose.

(*c*) The serum protein balance and the possible rôle of liver dysfunction.

(*d*) Examination of nasal smears was performed with the purpose of confirming the clinical diagnosis.

Results

1. *Nasal Smears:*

Contrary to the claims of most authors, the presence of eosinophils in smears from the nasal mucous membrane of nasal discharge was encountered only in a very small number of our cases, and even in those it was not a very marked finding (see Table I). However, nasal smears were a diagnostic aid in differentiating cases of infective rhinitis from pure allergic cases, the former showed a large excess of polymorphonuclear neutrophiles.

2. *Blood Eosinophils:*

More than half the cases of allergic rhinitis showed a definite increase in the blood eosinophils (see Table I). Cases that had parasitic infestation were excluded. The absence of local eosinophils in the nose and the presence of blood eosinophils could be explained by the assumption that the offending allergens in our cases were not inhalants but mostly ingestants. This assumption could explain the discrepancy between our findings (absence of nasal eosinophils) and the findings of other authors abroad. It is supposed that the offending allergen could affect the local formation of eosinophils at the site of action of the allergen on the mucous membrane. This suggestion had been experimentally supposed by Godlowski (1953).

No significant increase in the erythrocyte sedimentation rate was encountered in any of the cases of the allergic group.

TABLE I.

ALLERGIC GROUP.

Case No	Age	Sex	Duration	Nasal Smear	Total Leuc Count	Eosinophils %	Average Sed Rate per hour
1	25	M	6 years	Epithelial cells	7,200	5	7 mm
2	28	M	3 ,,	,,	4,200	4	6 ,,
3	22	F	6 ,,	,,	4,500	4	19 ,,
4	26	M	5 ,,	,,	5,600	2	5·5 ,,
5	25	M	10 ,,	Few epithelial cells	4,200	2	6 ,,
6	21	M	1 ,,	Rare eosinophils	4,200	6	6·5 ,,
7	21	M	3 ,,	Few epithelial cells	8,400	12	6·5 ,,
8	16	M	10 ,,	,,	7,600	11	15·5 ,,
9	17	F	3 ,,	,,	7,200	8	12 ,,
10	19	F	2 ,,	Rare eosinophils	6,000	9	11 ,,
11	16	F	3 ,,	Few epithelial cells	6,100	5	18 ,,
12	40	M	1 ,,	Rare eosinophils	5,200	8	3·5 ,,
13	30	M	3 ,,	Few epithelial cells	4,300	4	5·5 ,,
14	36	M	6 ,,	,,	8,200	14	7 ,,
15	32	M	1 ,,	,,	10,500	9	18 ,,
16	28	F	3 months	Rare eosinophils	9,300	10	14 ,,
17	26	F	3 years	Few epithelial cells	7,300	10	8 ,,
18	33	M	5 ,,	,,	5,800	8	6 ,,
19	40	F	4 ,,	,,	6,400	4	8 ,,
20	25	F	2 ,,	,,	8,200	5	7 ,,
21	18	M	5 ,,	,,	7,300	6	10 ,,
22	30	F	3 ,,	,,	6,800	5	11 ,,
23	33	F	2 ,,	,,	9,200	4	9 ,,
24	28	F	3 ,,	,,	8,400	6	8 ,,
25	26	M	4 ,,	,,	9,200	8	12 ,,

3. *Urinary 17 Ketosteroids:*

The values of urinary 17 ketosteroids ranged between 2·3 mg. and 8·0 mg. per 24 hours in the females and between 3·4 mg. and 10·0 mg. in the males. The figures were on the whole either on the low side of normal or slightly lower than normal. This is in contrast to the very low readings previously encountered in cases of genuine allergic bronchial asthma. It was observed that the cases that had very low 17 ketosteroids were found to be actually suffering from early manifestations of bronchial asthma too. Moreover, in pure infective rhinitis urinary 17 ketosteroids were found to be normal. The level of urinary 17 ketosteroids therefore might be taken as an early laboratory sign of change or progress of allergic rhinitis to bronchial asthma. The level of 17 ketosteroids would thus be of help in deciding the type of treatment. Good response to cortisone was observed in those cases with low excretion of the hormone. (See Table II.)

TABLE II.

Case No.	17 Ketosteroids Excretion/24 hrs.	Ser. Sod. M.E./litre	Ser. Pot. M.E./litre	Type of Treatment	Response to Treatment
1	8·4 mg.	142	4·2	Vitamin C	+
2	3·4 ,,	144	4·6	Cortisone	+++
3	7·2 ,,	148	4·2	Rutin	o
4	9·5 ,,	148	5·8	Calcium	o
5	9·0 ,,	146	4·2	Rutin	o
6	7·2 ,,	152	3·8	Antihistaminic	++
7	7·2 ,,	154	4·2	,,	++
8	5·9 ,,	146	4·4	Cortisone	+++
9	7·0 ,,	138	4·5	Antihistaminic	+
10	6·4 ,,	143	3·4	,,	+
11	2·3 ,,	143	5·0	Cortisone	++
12	7·2 ,,	148	4·0	Antihistaminic	o
13	5·6 ,,	149	4·0	Cortisone	++
14	10·0 ,,	148	4·2	Rutin	o
15	7·0 ,,	147	3·9	Cortisone	++
16	8·1 ,,	150	4·3	Calcium	o
17	8·5 ,,	145	4·2	,,	o
18	10·0 ,,	146	4·0	Rutin	o
19	9·8 ,,	146	4·1	,,	o
20	8·3 ,,	140	4·0	Vitamin C	o
21	8·6 ,,	146	5·0	Calcium	o
22	8·5 ,,	150	4·6	Vitamin C	+
23	8·3 ,,	143	4·7	,,	o
24	7·8 ,,	142	4·3	Calcium	o
25	7·0 ,,	149	4·4	Vitamin C	o

4. *Serum Protein Fractions:*

It was found that in almost all cases there was disturbance of normal serum protein balance. The Alb./Glob. ratio ranged between 0·58 and 1·7 instead of the normal 1·5 and 2·0. The disturbance was due to an increase in the globulin fraction in all cases. No diminution in the albumin level was observed in any case.

The main rise in the globulin fraction was due to an increase of Beta globulin in 20 out of 25 patients with allergic rhinitis. In 6 cases there was a concomitant increase of Gamma globulin too. In 3 cases the increase in the serum globulin was due to an increase in the Gamma fraction alone. (See Table III.)

Our finding of an increase in Beta globulin in allergic rhinitis, is in accord with other workers' finding (Raynaud *et al.* (1954)).

The same changes were encountered in other allergic conditions investigated on the same lines (Awny *et al.*, 1957). The outstanding rise in the Beta globulins could be explained, if we accept the enzymatic concept of allergy as the fundamental background of hypersensitivity. This theory (Godlowski, 1953) suggests that when a living cell is exposed to a foreign protein, the entrance of this protein into the cell would initiate the formation of a new specific toxic enzyme or the activation of the already existing cellular enzyme that makes it capable of destroying any further dose of this particular protein to toxic end products (polypeptides, etc) This formation of polypeptides will disturb the lipoproteins equilibrium in the cells to the extent that the cell proteins lose the protective action of lipoids against the cell enzymes This results in damage or autolysis of the cell protein, and a consequent release of lipoids These lipoids circulate in the blood in the form of lipoproteins, and might account for the rise in the beta globulin fraction in the serum protein

TABLE III

Case No	Ser Tot Proteins gm %	Alb	Globulin Fraction			Serum Glob gm %	Alb /Glob Ratio
			Alpha	Beta	Gamma		
1	8 4	4 1	2 3	0 64	1 36	4 3	0 95
2	9 2	4 1	1 6	1 18	2 32	5 1	0 80
3	9 8	4 5	1 4	2 14	1 66	5 3	0 85
4	10 1	5 2	0 8	2 98	1 12	4 9	1 06
5	9 4	3 9	1 3	2 88	1 32	5 5	0 70
6	10 0	4 7	1 0	2 86	1 48	5 3	0 90
7	9 0	4 6	1 2	1 84	1 36	4 4	1 04
8	10 6	4 1	1 4	3 62	1 48	6 5	0 63
9	10 3	4 2	2 1	2 00	2 00	6 1	0 70
10	9 0	4 1	1 6	1 42	1 88	4 9	0 83
11	9 8	4 2	2 1	1 82	1 68	5 6	0 75
12	9 4	4 6	1 5	1 66	1 64	4 8	0 96
13	5 8	4 2	2 3	0 54	1 46	4 3	0 98
14	8 2	5 2	0 7	0 70	1 60	3 0	1 70
15	8 6	4 2	2 3	0 74	1 36	4 4	0 96
16	9 6	3 5	1 3	1 70	3 10	6 1	0 58
17	7 2	4 2	0 9	1 20	0 90	3 0	1 40
18	8 4	3 9	1 4	1 50	1 00	4 5	0 87
19	9 8	4 8	2 2	1 68	1 12	5 0	0 96
20	9 0	4 7	1 2	2 20	0 90	4 3	1 01
21	9 8	3 5	0 8	2 70	1 80	5 3	0 68
22	8 6	4 1	1 0	1 80	1 70	4 5	0 90
23	9 2	4 7	0 9	2 40	1 20	4 5	1 00
24	8 5	4 8	0 7	1 90	1 10	3 7	1 30
25	10 2	3 8	1 52	2 40	2·48	6 4	0 60

5 *Changes in Electrolytes*

The relationship of different ions to various allergic diseases has received attention from several authors Rusk (1939) has found that the concentration of serum potassium is increased in various allergic states Donovan and Harsh (1939) failed to find any connection between allergic symptoms and serum sodium Later the same authors (1943) re-studied the problem on asthmatic children. They found that in these patients there was a high serum potassium and a low serum sodium. In addition, the asthmatic children were found to absorb from their intestines, and excrete in their urine, more sodium chloride than the normal controls. Sheldon *et al.* (1939) stated that prolonged asthmatic attacks were accompanied by urinary loss of sodium which may exceed the sodium intake by 3·0-5·0 grams per 24 hours. On the other hand Kern (1940) stated that sodium retention is a contributory factor in allergic manifestations. The same problem was also investigated on isolated preparation taking guinea pig's uterus response to histamine as an indicator. McDowall and Soliman (1954) found that histamine tachyphelaxis is abolished by replacing the physiological bathing solution of the preparation by one in which sodium chloride was reduced and potassium chloride was increased keeping osmotic pressure constant. This contradiction led us to investigate the changes in the electrolyte balance in allergic rhinitis. Our results indicated the existence of a slight but definite increase in the serum sodium, but no change in the serum potassium was found (see Table II).

In view of our present results we cannot decide the significance of this rise in serum sodium. We can, however, attain Kern's view that sodium retention consequently lead to water retention, which might contribute to the symptomatology of allergic rhinitis.

6. *Effect of Therapeutic Agents:*

Since the main symptomatology of the patients suffering from allergic rhinitis is generally a manifestation of increased permeability of the nasal mucosa, we expected to get relief of symptoms in these patients by administering drugs that might diminish the permeability of the capillary endothelium. We tried for this purpose (1) Vitamin C, (2) Vitamin P, (3) Calcium.

Vitamin C: It has been claimed by several authors that Vitamin C has a beneficial effect in allergic diseases. Parrot, Debray and Richet (1945) stated that ascorbic acid decreases the sensitivity of tissue to histamine, and also decreases the release of histamine during anaphylactic, peptone or allergic shock. Ungar (1944) demonstrated, on isolated preparations, that histamine release is significantly diminished if the experimental animal (guinea pig) would have had a dose of ascorbic acid beforehand. Again, Decourt and Parrot (1950) stated that Vitamin P (Vitamin C.2) augments the beneficial effect of Vitamin C against the injurious effect of histamine on isolated preparations. However, we used Vitamin C and Vitamin P for another important therapeutic effect which is their action in decreasing the permeability of capillary endothelium, assuming that allergic rhinitis has a permeability defect contributing at least partially in its pathogenesis In our work the dose of Vitamin C was 500 mg twice daily, for an adult of average 60 90 kilogram body weight The failure of Vitamin C in our cases might be explained by this wide difference in dosage

Calcium Therapy For a long time calcium has been used therapeutically in the treatment of diverse clinical manifestations including allergic ailments It was an everyday experience to prescribe calcium for patients with allergic rhinitis, bronchial asthma, eczema and urticaria The idea was that, calcium ions have a sedative effect on nerve endings and also decrease the permeability of blood vessel walls Experimentally it has been shown by certain authors that calcium ions counteract the effect of histamine on isolated preparations Not only this, but Halpern (1945) has shown a relationship between antihistamine–histamine antagonism and calcium -ion concentration in the bathing fluid of an isolated preparation In the present work we used calcium gluconate (1 gram intravenously daily for at least two weeks) without any beneficial effect In view of the tremendous literature on this item, and our results we do not feel justified in advising calcium in the therapy of allergic rhinitis

Considering the assumption of histamine release in the pathogenesis of symptoms in various allergic states, we tried the effect of antihistamine drugs We have chosen an antihistaminic in which both the sedative and anti-acetyl choline effect were minimal, namely mepyramine maleate (Gaddum, 1948) Its action is mainly antihistaminic The result was encouraging This is in contrast to the well known failure of antihistaminic therapy in bronchial asthma The beneficial effect of antihistaminics in our present work gives a partial support to the histamine release theory in allergic rhinitis

Cortisone Therapy The suprarenal gland has ever been claimed to have a definite rôle in hypersensitivity states, in histamine shock, allergic reactions, and various stress syndromes Feldberg and Schelf (1930), in their book on histamine, have demonstrated the effect of suprarenal medulla on the resistance of intact animals to various shock syndromes No doubt it is the adrenaline of the suprarenal medulla that has the effect of raising the resistance of animals against stress Later it has been found that the adrenal cortex also has a major rôle in stress syndromes Evidence of general depletion of the suprarenal cortex was demonstrated in the severe cases of stress syndromes It has also been found by several authors that cortisone has a marked and rapid beneficial effect in certain allergic states

Several interesting explanations for the mode of action of cortisone in allergic phenomena were given, among which could be mentioned

(1) Cortisone potentiates the formation of hormonal antibodies and

therefore counteracts the effect of antigen on cellular antibodies and consequently prevents the sensitization shock.

(2) Cortisone potentiates the action of histaminase and therefore counteracts the effect of histamine released after the combination of antigen with the antibody, Kahlson (1956).

(3) Cortisone re-arranges the enzymatic discord claimed to be the basis of allergic manifestations.

(4) Cortisone has an anti-hyaluronidase effect.

(5) Cortisone has an effect over the permeability of blood vessels and also on collagen fibres and hence its beneficial effect on the collagen diseases.

In this study we used Ultracorten (Ciba) tablets each containing 5 mg. The dosage used was the minimum amount capable of maintaining clinical improvement. We found that a course starting with 5 mg. 6-hourly for 3 days followed by 5 mg. 8-hourly for 5 days is quite satisfactory. This was followed by a gradual withdrawal of the drug. Complete or almost complete relief of symptoms was obtained in all cases for which this treatment was used. (See Table II.)

Summary and Conclusion

The results of certain laboratory studies directed towards the detection of possible disturbances in endocrine, electrolyte and protein balance associated with a mild genuine allergic state (allergic rhinitis) are discussed. A definite disturbance of the serum protein fractions manifested mainly by an increase of the Beta globulin fraction was encountered in almost all cases. Certain cases showed a concomitant increase in the gamma globulin fraction too. A slight elevation of the serum sodium level was observed. Definite functional disturbance of the suprarenal cortex was present in advanced cases. Local eosinophilia of the nasal mucosa and of the nasal discharge were almost absent while blood eosinophilia was present in most of the cases. The relation of these findings to the nature of the offending allergens was discussed.

A comparison of the effect of three different types of treatment on the symptomatology of the condition was included in the study. The best results were obtained through the action of cortisone. Antihistaminics were less effective than cortisone, while drugs desired to diminish the capillary permeability failed to give any relief.

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CLINICAL RECORDS

CHOLESTEATOMA OF THE MAXILLARY ANTRUM

By C. PETER MILLS and E. McK. SYCAMORE (London)

CHOLESTEATOMA of the maxillary sinuses is rare. The following case is reported because of its close clinical similarity to a carcinoma of the antrum.

Case Report

Mrs. E. H., 65 years, complained of pain over the left cheek associated with a foul tasting discharge from a sinus in the left upper alveolar region.

June, 1957—the patient was seen by a dental surgeon having had a persistent left rhinorrhœa, some left nasal obstruction and pain in the left cheek and frontal region for one year. She had had treatment for maxillary sinusitis with a variety of antibiotics and nasal drops without effect; the pain was not alleviated by simple analgesics.

Examination at that time revealed a sinus near the root of the |6. The |6, 7 were extracted. She was seen again by her dentist in August complaining of a more severe pain and swelling over the region of the |7. and a foul discharge into the mouth. Excessive granulations were observed in the region; she was therefore referred to hospital.

August 29th—there had been a swelling over the left cheek for three weeks. There was a large oroantral fistula at the site of |6, 7. which exuded foul pus; a silver probe passed easily into the antrum. Expansion of the left alveolus with a deficiency of the lateral wall was noted. An X-ray (Fig. 1) showed an opacity of the left antrum with extensive destruction of the floor, medial and lateral walls.

Some of the exuberant granulation tissue from around the fistula was removed for histological examination, this showed œdematous granulation tissue covered partly with squamous and partly with respiratory type epithelium. Despite the negative histological findings it was felt that clinically this was a carcinoma of the antrum.

September 3rd. Excision biopsy of the fistula under general anæsthesia.

Both fistulae and $\frac{1}{2}$ cm. of surrounding healthy alveolar epithelium were excised. Through the resulting hole the antrum was seen to be full of a foul smelling whitish mass. It was removed with the aid of suction and appeared to be made up of sheets of material similar to that seen in a cholesteatoma of the mastoid. Bleeding was slight. There was no macroscopic evidence of a neoplasm. Much of the lateral, medial, and anterior walls was felt to be absent. The fistulous area, and some antral mucosa were sent for histological examination. It was reported that there were inflammatory polypi covered by innocent columnar epithelium which showed metaplasia in some parts (Fig. 2). No evidence of malignant change could be found, no cholesterol crystals were seen.

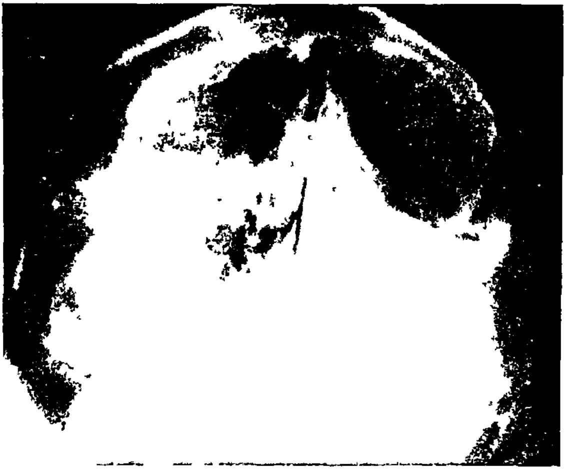

FIG 1

Shows opacity of the left antrum with destruction of the floor medial and lateral walls

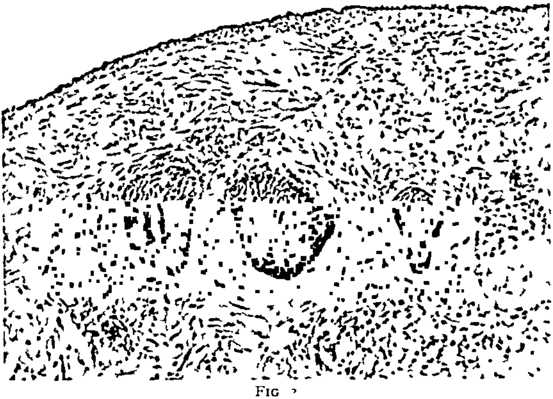

FIG 2

Shows squamous metaplasia in the lining epithelium of the antrum ×135

Following drainage and clearance of the antrum the pain was relieved, the patient feeling very much better.

September 19th. Under general anæsthesia the edges of the fistula were excised, and the palatal mucosa was mobilized, and a small amount of bone was removed from the palate and maxillary tuberosity. The antral lining which was thick and leathery was removed *viâ* the fistula. A large nasal antrostomy was produced, the fistula was then closed without tension by interrupted mersutures.

The patient was discharged two weeks later, the wound having healed by first intention.

Ætiology

The exact pathogenesis of aural cholesteatomata is uncertain, most authorities agree that infection is or has been present at some time, and that squamous epithelium is an essential precursor.

In propounding a theory on the ætiology of cholesteatoma of the maxillary antrum, Petrillo (1949) suggested that it may develop through:

(1) Invasion of the sinus cavity by buccal epithelium after trauma *viâ* an antro-alveolar fistula.

(2) Inclusion of the epidermis in the sinus cavity during fœtal life.

(3) Metaplasia (rare).

Some authorities, Pinus (1949) and Shcherbatoff (1953), believe that the vast majority of cases occur in association with dental cysts that obliterate the maxillary antrum and are filled with cholesteatomatous material. It is interesting to note that Shcherbatoff was able to find 21 cases of cholesteatoma of the maxillary antrum over a period of two years in one hospital, yet there are fewer cases recorded in the entire world literature over the past fifteen years. If the majority of cholesteatoma were due to dental cysts filling the antrum one would expect expansion of the antrum, rather than actual destruction of the walls.

A dental origin cannot be blamed for those found in the frontal sinuses, which are far commoner than antral cases.

It is therefore suggested that a true cholesteatoma of the antrum is secondary to chronic antritis which has given rise to a squamous metaplasia, this epithelium being the precursor of the cholesteatoma.

Discussion

Hutcheon (1941) in describing a case "of rhinolith combined with cholesteatoma of the maxillary antrum", said "the most interesting feature of this case is that the symptoms and signs were much more suggestive of carcinoma of the antrum and adjacent nasal wall than of any other diagnosis". A condition which may be confused with cholesteatoma of the antrum is caseosis. In these cases a mass of foul smelling caseous material is associated with a shaggy necrotic antral lining but no bone destruction, whereas with a cholesteatoma there is bone destruction with little or no expansion of the walls, the contents of the antrum being in sheets of whitish material like the layers of an onion.

Treatment

A complete cure may be obtained by stripping all antral musosa, and making a large nasal antrostomy for drainage.

Summary

1. A case of cholesteatoma of the maxillary antrum is described.
2. A brief review of its possible ætiology is made.

Acknowledgements

We wish to thank Mr. Philip Reading for his permission to publish this case, and for his kind help in the preparation of this paper Our thanks are also due to the Dental department, Guy's Hospital, for the X-ray and photomicrograph.

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AN UNUSUAL CASE OF UNILATERAL NASAL POLYPI

By J. D. K. DAWES (Newcastle)

THIS case history is recorded as being of interest.

E.S., æt 35. This patient was first seen at Berwick Infirmary on *October 31st, 1955*, complaining of right-sided nasal obstruction. On examination the right nasal cavity contained polypi arising mainly from the middle and posterior ethmoid cells. In the post-nasal space a little pus could be seen in the right choana.

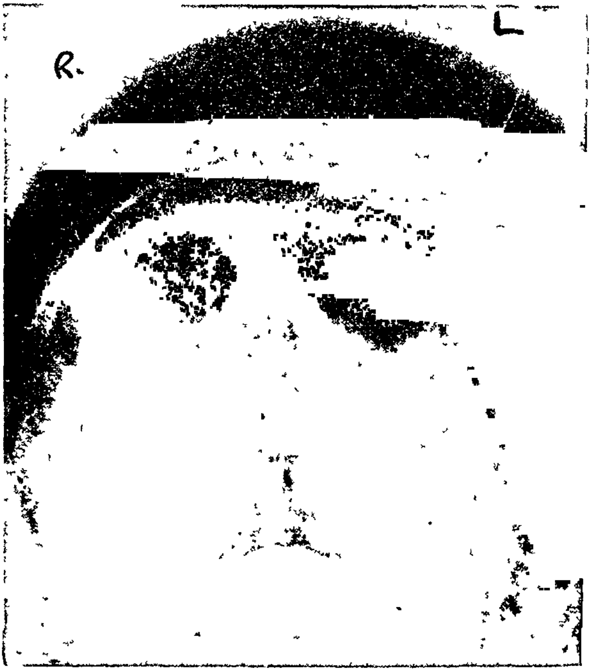

FIG 1
Radiograph showing a foreign body in the right antrum.

On *November 7th, 1955*, the polypi were removed and a large cavity in the right posterior ethmoids was opened and found to be filled by masses of black dust which had become muddy in consistency and covered by a layer of inspissated pus. A wash out of the right maxillary antrum produced further masses of black dust which the patient instantly recognized as "brass dust", a colloquial term for iron pyrites which is used in the manufacture of acid His normal occupation was to shovel the iron pyrites into the furnaces

The patient was again seen on *November 21st, 1955* He still complained of a right-sided nasal obstruction although he was free of polypi The nasal mucosa was seen to be very œdematous An X ray of sinuses showed the presence of a foreign body in the right maxillary antrum (Figs 1 and 2)

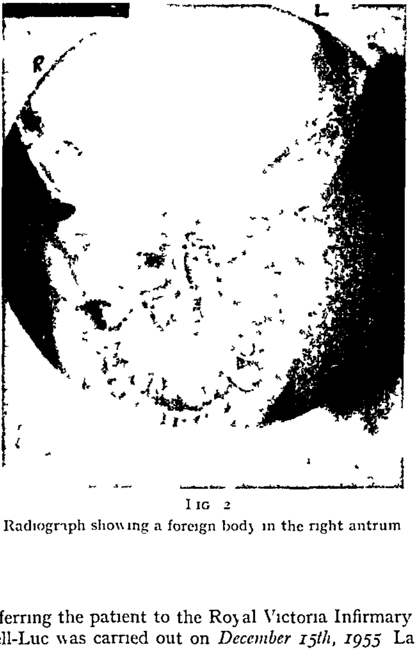

Fig 2

Radiograph showing a foreign body in the right antrum

After transferring the patient to the Royal Victoria Infirmary in Newcastle, a right Caldwell-Luc was carried out on *December 15th, 1955* Large chunks of black dust filled the maxillary antrum A large foreign body (Fig 3) covered by the dust was found on the floor of the antrum The antral mucosa was thickened and a specimen sent for histopathological studies The whole antral wall above the left inferior turbinate was missing and the posterior ethmoid region consisted of a single large cavity in direct communication with the maxillary antrum After creating a nasal antrostomy below the level of the inferior turbinate the buccal wound was sutured On *December 21st, 1955*, the buccal wound was beginning to slough and the patient was given Distaquaine Penicillin 600,000 units twice daily for 8 days The buccal wound healed very rapidly after this

The patient was last seen in *October, 1957*; he now has no complaints and has returned to his previous occupation.

22.12.55. Pathology Report. (*Dr. R. Schade.*)

Histologically the antral mucosa shows œdema and intrinsic chronic inflammatory changes as well as foreign bodies which give a slightly positive iron reaction.

12.4.56. Biochemical Report. (*Dr. A. L. Latner.*)

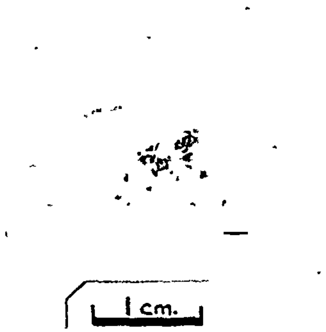

FIG 3.

Foreign body extracted from the right antrum.

The black powder is iron sulphide. The central hard core (Fig. 3) consists mainly of iron with an outer coating of iron sulphide. In my opinion this could be a piece of shrapnel and the outer coating of sulphide could have been produced by bacterial action. I do not think this is a lump of iron pyrites.

In retrospect the patient was questioned more closely regarding shrapnel wounds. He was a German who had settled in the area after being a prisoner of war. A shrapnel wound had been received on the Russian front in 1943, the wound of entry being beneath the left mastoid process. Soon after the wounding, small pieces of shrapnel had been removed and the wound healed satisfactorily.

Comment

It is only in retrospect that the presence of a metallic foreign body in the right maxillary antrum was found to be shrapnel which presumably had traversed the post-nasal space before entering the antrum, since there was no obvious damage to the nasal structures. Secondly, he developed nasal polypi as a consequence of irritation by iron pyrites either of occupational origin or possibly, as Dr. Latner suggests, by the effects of bacterial action on the shrapnel.

SOCIETIES' PROCEEDINGS

ROYAL SOCIETY OF MEDICINE—SECTION OF LARYNGOLOGY

President R J CANN, M S, F R C S

December 6th, 1957

Discussion on Pharyngeal Diverticula

THE opening papers by Mr Boyes Korkis and Mr Spencer Harrison will be found on pp. 509 534

Discussion

PROFESSOR E W WALLS opened the meeting with a brief account of the anatomy of the lower pharynx with particular reference to the innervation of the inferior constrictor

SIR VICTOR NEGUS thanked the President for inviting him to speak He had a long-standing interest in pharyngeal diverticula, but he would not weary the meeting with any remarks about etiology based on the evidence of comparative anatomy, since he had published articles in the *Journal of Laryngology* in 1925, in the report of a lecture in the *Annals of the College of Surgeons* in 1950 and recently the report of a lecture given at the Johns Hopkins Hospital, published in the Bulletin of that institute

His opinion was that a pharyngeal diverticulum, unless extremely large, was the result and not the cause of dysphagia, it was usual to find evidence of lack of relaxation of the cricopharyngeal sphincter, sometimes associated with chronic inflammatory changes By means of dilatation with a large hydrostatic bag it had been found possible to make certain small pouches disappear, with relief of all symptoms, and in one case, of a moderate sized pouch, preliminary dilatation was to be followed by external operation after some days, but when this was arranged it was found that there was no evidence of a pouch

Sir Victor said that he knew of the operation proposed by Dr Mosher many years ago, with division of the party wall as far down as within a quarter of an inch of its lower margin and without diathermy This had appeared to be somewhat precarious, but the recent improvements in the operation introduced by Dr Dohlman seem to have made the procedure a safe one Dr Seiffert of Berlin had, it was believed, carried out similar operations

W O LODGE asked if the openers had found themselves, during excision of pharyngeal diverticula, practising *pari passu* repair of the pharynx as shown in a diagram , see Fig 1

E D D DAVIS said he had seen about 26 cases of pharyngeal diverticula during a period of 35 years and he had collected brief notes on 19 of them

X-ray photographs were sometimes misleading and showed the fundus of the diverticulum to be below the level of the clavicle and in the mediastinum, whereas in all the 19 operations he had seen the fundus was in the neck well above the clavicle.

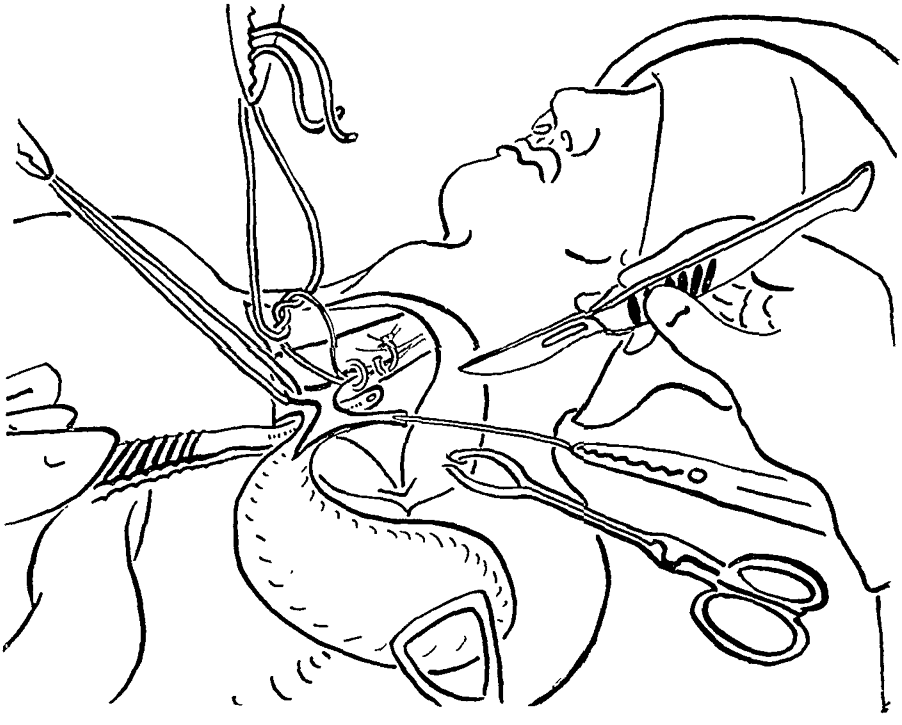

FIG. 1.

Endoscopy was always necessary before operation and it helped to determine whether the diverticulum was suitable for the Dohlman operation or for excision. In his day sepsis and mediastinitis were the bogies of operation and several methods of treatment were tried to avoid these complications. The first case he saw was submitted to Gerard's operation, i.e. the diverticulum was exposed by an oblique incision along the anterior border of the sternomastoid, the omohyoid muscle was divided, the pouch dissected and invaginated into the pharynx. The slit-like opening left in the wall of the pharynx was carefully closed by stitches. During the operation, before the closure of the slit, the patient coughed, with the result that the diverticulum returned to the position in the neck. He was made to cough after stitching, and the diverticulum remained securely in the pharynx. Two months later an attempt was made to remove the polyp-like diverticulum with the œsophagoscope. The patient disappeared for nearly 12 months, when he reported that he had sneezed violently, felt something give in his neck and his symptoms returned. The diverticulum was shown by radiography to be in the same position as before operation It was then excised at a second operation, but a slight recurrence was seen 18 months later

The next four cases were treated by what is known as diverticulopexy The fundus of the diverticulum was drawn up in the neck and stitched to the hyoid bone Three of the diverticula returned to their original position in the neck within two months A fourth sloughed and a fistula resulted In his experience the two stage operation resulted in a return of the diverticulum He also knew of two such cases who died of mediastinitis

The one stage operation done by Wilfred Trotter was the most successful After the diverticulum was excised the pharyngeal wall was closed by four layers of sutures The sternal portion of the sternomastoid muscle was divided at the sternum and the muscle stitched over the carotid sheath to fill up the gap and to strengthen the pharyngeal wall Trotter treated the diverticulum as a hernia which he considered to be caused by the increased pressure within the pharynx arising from the functional contraction of the cricopharyngeus Dunhill's collar or transverse incision, done for æsthetic reasons, resulted in injury to the recurrent laryngeal nerve in one case There was a recurrence in a few of the excision cases

Wednesday, 2nd July, 1958, at 5.30 p.m.
Dr. Enrique Ager Muguerza (Madrid)—"Carcinoma of the epiglottis."

Thursday, 3rd July, 1958, at 5.30 p.m., at The Royal College of Surgeons.
Mr. Norman Tanner—"Hæmorrhage from the œsophagus."

Friday, 11th July, 1958, 4.0 to 7.30 p.m.
Reception and demonstrations in the Audiology Unit.

Friday, 8th August, 1958, at 5.30 p.m. (instead of 30th July as previously notified).
Dr. Ben H. Senturia (St. Louis)—"Diseases of the external ear."

Friday, 24th October, 1958, at 5.30 p.m.
Professor C. V. Harrison (Postgraduate Medical School of London)—"The pathology of malignant lymphomas."

Saturday, 8th November, 1958, at 11.30 a.m.
Professor Lucius Rüedi (Zurich)—"The modern treatment of cholesteatoma of the middle-ear cleft."

Friday, 14th November, 1958, at 4.30 p.m.
Mr. T. G. Wilson (Dublin)—ANNUAL ADDRESS.

Friday, 12th December, 1958, at 5.30 p.m.
Professor W. St. C. Symmers (Charing Cross Hospital Medical School)—"Mycotic diseases".

Provisional List of
Short Courses for Consultants and Senior Registrars

3rd and 4th October, 1958—Practical Aural Surgery Class.

17th, 18th and 19th February, 1959—Malignant diseases of the ear, nose and throat.

21st and 22nd February, 1959—Pathology of the ear, nose and throat.

10th, 11th and 12th March, 1959—Diagnosis and treatment of the deaf child.

13th and 14th March, 1959—Modern methods of acoustic and vestibular testing.

10th and 11th April, 1959—Practical Aural Surgery Class.

The number of entries is limited in each course.

A detailed syllabus of any of the above can be obtained on application to the Secretary.

The Journal of
Laryngology and Otology

(*Founded in* 1887 *by* MORELL MACKENZIE *and* NORRIS WOLFENDEN)

August 1958

CHOLESTEATOSIS OF THE ATTIC*

By L RUEDI (Zurich)

I FEEL greatly honoured to have been asked to address you on the old subject of cholesteatomatous otitis media I must confess that I am not sufficiently familiar with the term "cholesteatosis of the attic" to be able to give it a precise definition

We differentiate between chronic otitis media with a central perforation and that with an upper marginal perforation where the tympanic defect leads into the attic Cholesteatomas occur as a rule only in the latter type of middle-ear infection and the cholesteatomatous lamellae in the attic can be seen through the upper marginal perforation Cholesteatomas in the middle ear may be classified into two groups according to the extent of the perforation The larger of these includes cases with *large* upper marginal perforations, while the smaller group includes the less common cases with *small* upper anterior or posterior marginal perforations In 1934 statistical analysis of 763 cases of cholesteatomatous otitis media seen at the Zurich ear clinic placed 631 cases into the first category with large upper marginal drum defects, while only 132 cases fell into the second category with small marginal perforations

Intracranial complications occurred in only 1 7 per cent of the larger group of cholesteatomas with *extensive* drum defects While the classical radical operation obviates this danger, the unpleasant otorrhoea tends to continue in many patients, and hearing often deteriorates after the radical mastoidectomy. These unsatisfactory results have prompted many otologists to adopt conservative methods in the management of these cases. The large tympanic defects enables one to reach the cholesteatoma relatively easily and to remove the masses of dead epithelium from the middle ear. As a result of this the discharge from the ear usually stops, but, of course conservative treatment cannot hope to improve the hearing. At best the patient will remain under medical supervision for the rest of his life.

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* Address, delivered by invitation, to the Section of Otology Royal Society of Medicine 7th February, 1958

In the smaller group of cholesteatomas with *small* upper marginal perforations the incidence of intracranial complications is four times higher (6 per cent. as compared with 1·7 per cent.). Conservative treatment cannot reach the affected area through the small drum defect and the danger of intracranial complication continues. Radical mastoidectomy is, therefore, the treatment of choice for these cases and has been recognized as such since the days of H. Schwartze, L. Stacke, K. Wittmaack *et al.* The institution of early surgery is amply justified by results. A conservative radical operation not only eradicates that form of cholesteatoma and its inherent danger to life, it will also preserve or even improve hearing and, as a rule, halt the unpleasant discharge from the ear.

It is to be expected that the technical advances of temporal bone surgery, and especially fenestration surgery, should lead to better results in the treatment of all forms of cholesteatoma. However, an exact knowledge of the pathogenesis of cholesteatomas is essential for the successful application of these new surgical techniques. In the Zürich series of cholesteatomas with large drum erosions, the middle-ear suppuration started as a necrotizing otitis media of infancy in 35 per cent. of cases. This arose in the course of scarlet fever, measles, diphtheria, tuberculosis or influenza. In the remaining 65 per cent. of cholesteatoma with large perforations no such history could be obtained. But in the majority of cases the middle-ear infection dated from early childhood and it may be assumed that these cholesteatomas also began as a necrotizing otitis media of unknown ætiology in infancy. J. Habermann was the first to observe the reaction of the epithelium of the external auditory meatus abutting on the attic perforation during an attack of necrotizing otitis media in infancy. As a result of this reaction meatal epithelium grows inwards through the perforation and once in the middle ear stimulates the production of a cholesteatoma. Some supporters of this immigration theory regard this ingrowing of the meatal epithelium as an attempt to repair the destroyed mucosa of the middle ear. P. Manasse has pointed out, however, that the cholesteatomatous matrix shows a strong tendency to proliferation, which is unlike the simple process of epithelial covering of the cavity. The invading squamous epithelium is not content to advance on the surface. It spreads mainly by sending forth strands and processes into the submucous connective tissue of the still intact cubical epithelium of the middle ear. Thus from its very beginning the development of cholesteatoma is not to be looked upon as a replacement of the destroyed mucosa of the middle ear by squamous epithelium. Rather is it to be seen as a struggle between the two types of epithelium. This struggle is rather an unequal one, Fig. 1, the cholesteatoma undermines the mucosal cubical epithelium, lifting it off the underlying connective tissue and pushing it away as it advances. The cholesteatomatous process frequently breaks through the intact mucosa

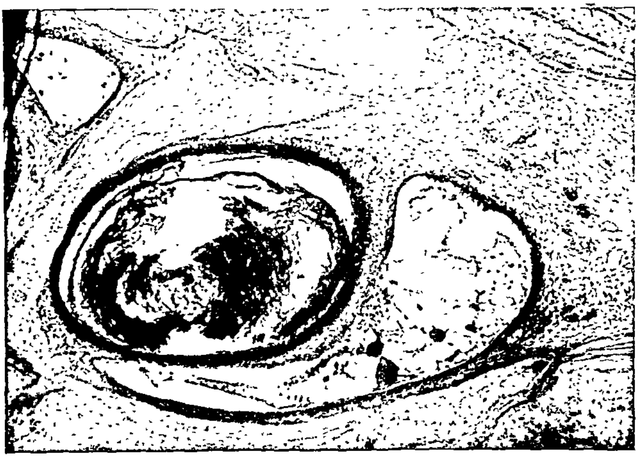

Fig. 1.

Otitis media chronica cholesteatomatosa in the case of an 11 year old boy. The cholesteatoma undermines the mucosal cubical epithelium

of the middle ear and (Fig. 2) once on the surface masses of desquamated squamous epithelium are shed into the cellular system. The mucosa reacts to this by producing an inflammatory exudate. The single layered cubical epithelium becomes heaped up into cushions many layers thick. The aggressive behaviour of the cholesteatomatous matrix in relation to the mucosa in cases with a large drum defect cannot therefore, be regarded as a simple harmless attempt at healing. On the other hand the matrix does not attack the surrounding bone (Fig. 3). In the early stages of the disease the bony tissues respond to the inflammatory stimulus by laying down new bone and by ossifying the submucous connective tissue. The active growth of the matrix in the depths of the submucosa, proceeds in the same connective tissue plane alongside the osteogenetic process, without either process interfering with the other. When all the available submucous connective tissue has been used up, the cholesteatomatous

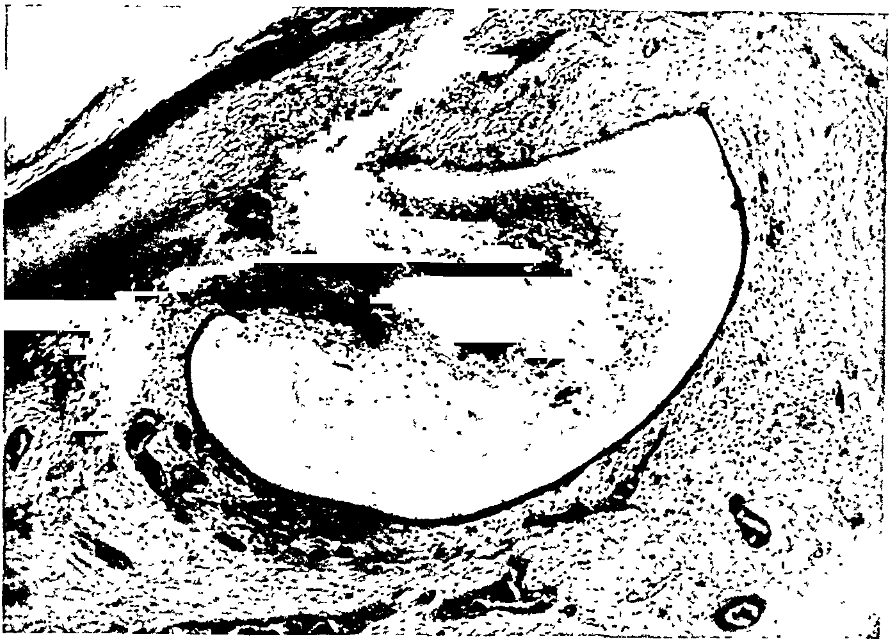

FIG. 2.
Otitis media chronica cholesteatomatosa in the case of an 11 year old boy. The cholesteatoma breaks through the mucosa of the inner ear.

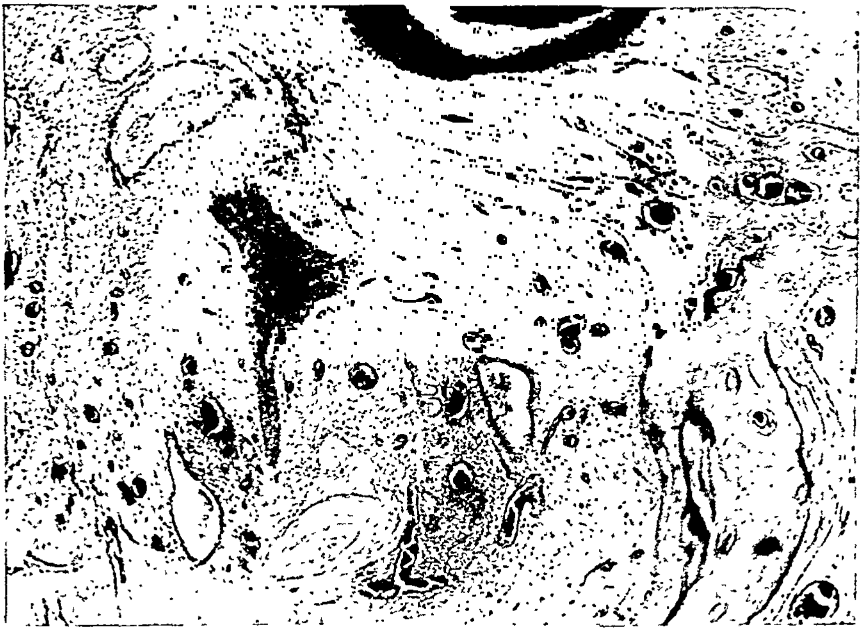

FIG. 3.
Otitis media chronica cholesteatomatosa in the case of a 23 year old man. The active growth of the matrix proceeds in the connective tissue alongside the osteogenetic process taking place in the same connective tissue.

channels become choked with desquamated squamous epithelium. It is only at this stage that the bed of the cholesteatoma begins to enlarge passively, the secondarily laid down sclerotic bone being reabsorbed as a result of direct pressure by the mass of the cholesteatoma. The passive expansion at the expense of surrounding bone, which is characteristic of the advanced cholesteatoma, is less important for the understanding of the whole disease than the early active growth of cholesteatomatous processes within the submucous connective tissue. There can be little doubt from the above observations that cholesteatomas with large tympanic perforations arise as a result of an inflammatory reaction causing immigration of the meatal epithelium.

On the other hand there is no unanimity of views concerning the pathogenesis of the rarer cholesteatomas with *small* upper marginal perforations. In the Zürich series there were 132 such cases and only about 20 per cent. gave a history of otitis media in early childhood. In none of the cases did an ear specialist observe the appearance of a small upper marginal perforation in the course of the otitis media. At the onset the disease is usually symptomless, without an obvious association of a middle ear infection. The small perforation is often discovered accidentally in the course of a routine examination. The character of the perforation at this early stage already shows signs of a cholesteatoma having become established to it. Operation on such early cases reveals a surprisingly large cholesteatoma in the attic. It is tempting to compare these cases with the very rare cholesteatomas with an intact ear drum, which are occasionally encountered during operations for other aural conditions.

Regarding this possibility M. Diamant states that "the invasion of the stratified squamous epithelium from the auditory meatus is impossible". He considers that these cholesteatomas arise, as suggested by R. W. Teed, from embryonic squamous epithelial remnants in the mucosa of the middle ear. Dan McKenzie speaks of "a primary tumour, similar to intracranial cholesteatoma", while as early as 1925 F. R. Nager pointed out that "the true cholesteatomas are so rare as to be negligible". The almost symptomless origin of some cholesteatomas is explained in the Anglo-Saxon literature by postulating a metaplasia of the mucosa of the middle ear to a keratinizing stratified squamous epithelium as a result of inflammatory changes. R. R. Simpson considers that this metaplasia may be induced by the formation of a cholesterol granuloma in the middle ear and his views are shared by A. Tumarkin.

Can the pathogenesis of these cholesteatomas of insidious onset and with small upper marginal perforations be explained on the basis of the immigration theory?

F. Bezold's hypothesis of a chronic tubal catarrh leading to retraction and finally rupture of Shrapnell's membrane finds little support today. While retraction of the pars flaccida is a common occurrence, its rupture *with consequent cholesteatoma formation has never been observed to the* best of our knowledge. There is no reason to suppose that such a perforation would not heal rather than lead to a cholesteatoma. F. Bezold himself was aware of this difficulty and pointed to the co-existing inflammatory factors as maintaining the perforation in Shapnell's membrane. P. Manasse drew attention to the proliferative powers of the drum around small, upper marginal perforations in the presence of chronic or subacute infection. J. Habermann considered this a possible factor in the aetiology of the cholesteatomas. Finally, W. Lange furnished histological proof of deeply ingrowing prickle cells in the epidermis of Shrapnell's membrane which was neither indrawn nor perforated. He considered that this epithelial proliferation was caused by an inflammatory stimulus such as a prolonged attack of otitis media. In W. Lange's opinion these proliferating columns of basal cells grow into the submucous connective tissue of Prussak's space, thus forming the basis of the cholesteatomas. W. Albrecht and M. Schwarz, have pointed to the importance of connective tissue rests in the epitympanic space in the formation of Shrapnell's cholesteatomas. The development of the middle-ear cleft proceeds in two stages. As we could show, in a first stage bone cavities are formed; these cavities are pneumatized in a second stage. We have also been able to show in 1937 and in 1939 that in many new-born and very young infants extensive deposits

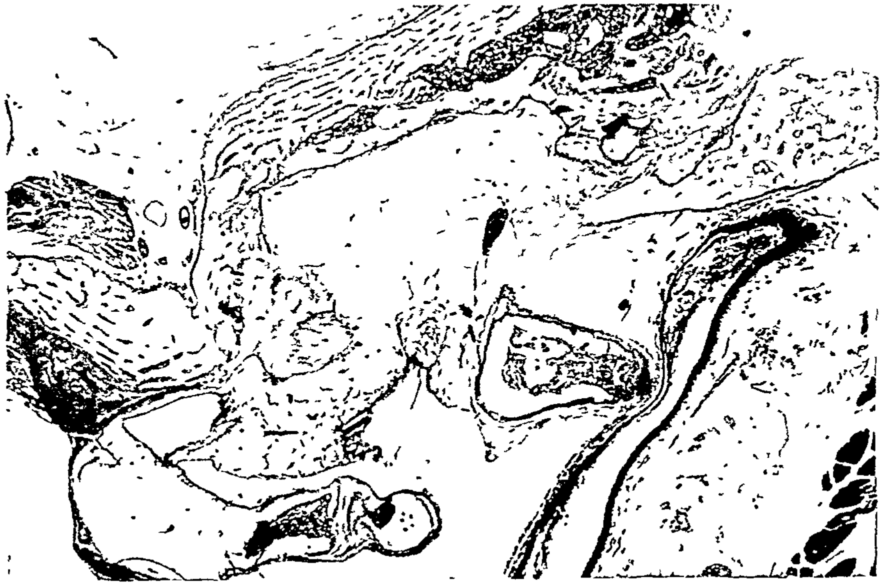

FIG 4

A vertical section through the middle ear of a new-born Submucous connective tissue fills the incompletely pneumatized attic of submucous connective tissue fill the incompletely pneumatized attic and epitympanic cells (Fig 4) Infants with this congenital "mucosal hyperplasia" are particularly liable to attacks of acute catarrhal inflammation of the middle ear This recurrent inflammatory process may stimulate the active proliferation of basal cells in the pars flaccida of the drum as suggested by W Lange It may also facilitate the growth of columns of basal cells into the depths of the submucous connective tissue in the attic and thus initiate a cholesteatoma

At the International Congress in Washington, D C (1957) I mentioned some observations which support W Lange's view

(1) The structure of the matrix of cholesteatoma with large drum defects and those with small upper marginal perforations are histologically identical with stratified squamous epithelium of the skin

(2) The matrix of both types of cholesteatomas has the same tendency to active invasive growth, while the mucosa, submucosa and bone of the middle ear react to this growth in much the same way in either case

(3) In the normal human fœtus of 7-9 months and in very young infants, the upper drum margin shows regularly a greatly augmented epidermis The growth of the epidermis at the junction with the external auditory meatus amounts to a hyperkeratosis and acanthosis

(4) As a result of low grade inflammation in the middle ear the growth of the meatal skin is activated These findings imply that cholesteatomas with small upper marginal perforations as well as with large marginal defects of the tympanic membrane originate by immigration of squamous epithelium from the drum or the auditory meatus

Differences in the early development, growth and the risks of complications in both types of cholesteatoma depend on the intensity of the inflammatory stimulus In cases with large drum defects, arising in the course of a necrotizing otitis media in infancy, the skin of the auditory meatus receives a powerful stimulus The squamous epithelium grows quickly through the large perforaton to invade the submucosa of the middle ear This active growth is, however, confined to soft tissues and does not attack bone The bone itself responds to the strong inflammatory stimulus by laying down protective bony lamellae and by a rapid ossifica tion of the submucous connective tissue The actively growing cholesteatomas are thus soon deprived of their mesenchymal basis The reinforced sclerotic bony walls (Fig 5) protect the internal ear and the endocranium at least as long as the dead cholesteatomatous material is extruded either spontaneously or therapeutically through the large tympanic perforation

In Shrapnell's cholesteatomas, however, the initial growth of the basal cells from the still intact pars flaccida is probably much slower and intermittent This corresponds to the much weaker stimulus of recurrent catarrhal middle-ear inflammations in early childhood. The slowly growing

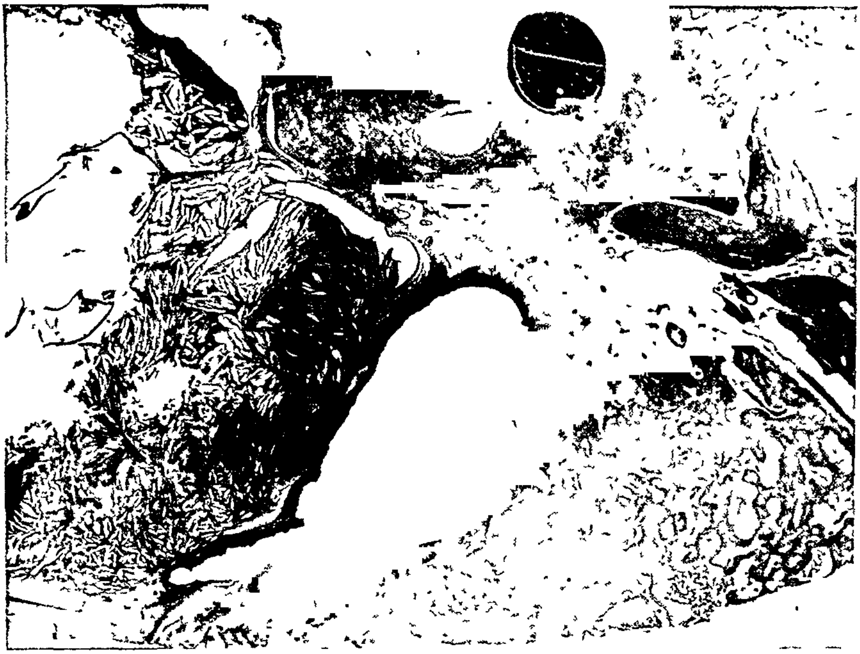

FIG 5
Otitis media chronica cholesteatomatosa in the case of a 35 year old man. The cholesteatomatous matrix is lining a wall of sclerotic bone.

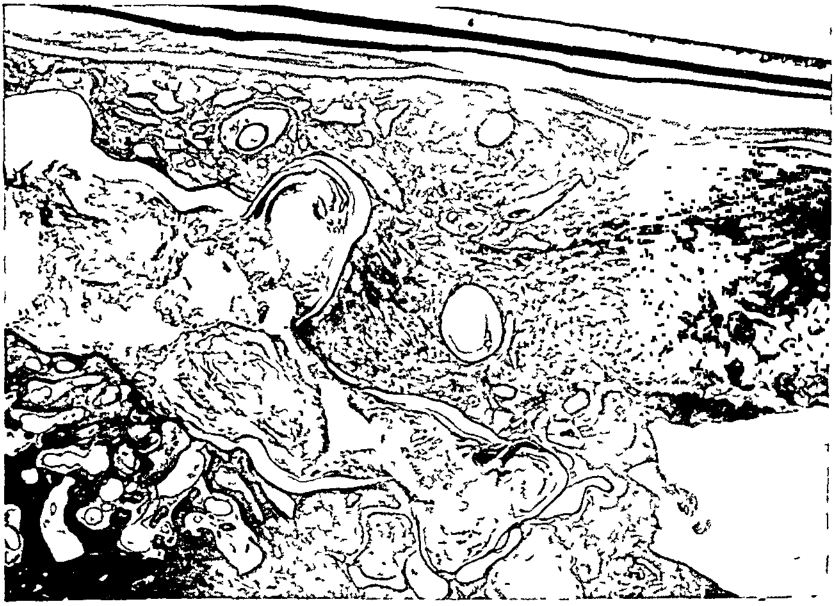

FIG 6
Shrapnell's cholesteatoma in the case of a not-operated 18 year old boy.

fingers of the early Shrapnell's cholesteatoma, nevertheless, extend deeply into the abundant connective tissue of the attic, while the drum remains intact As the bony reaction is also mild in these cases the connective tissues become only slowly ossified Long tubes of cholesteatoma grow out to the not yet pneumatized cell tissues before the appearance of a perforation By the time a small, upper marginal perforation forms, the disease is too extensive to be managed conservatively Having finally used up all the available submucous connective tissue (Fig 6), the cholesteatoma fills the narrow spaces around the attic, becomes infected and causes a pressure absorption of the neighbouring bone The infection cannot be reached through the small perforation and will continue to extend into the cranium Thus cholesteatomas with small upper marginal perforations are more dangerous by virtue of the local conditions which facilitate the deep penetration of the matrix while diminishing the protective reaction of the surrounding bone

One hundred and twenty four temporal bones obtained from cases of cholesteatoma, acute otitis media and from normal young children were examined histologically by serial section A search for evidence of embryonic cell rests or areas of metaplasia in the mucosa of the middle ear proved fruitless While the occurrence of such cell rests or metaplasia is possible, it is at best very rare, while cholesteatomas are encountered commonly

The suggestion was made by English workers that cholesteatomas may arise as a result of cholesterol granulomas A Tumarkin aptly observes that "the relation of cholesterol granuloma to cholesteatoma has given rise to much argument and the problem has been confused by a failure to define terms" The confusion in fact arises from the conflicting clinical terms applied to the histologically well-defined cholesterol granuloma G E Shambaugh Jr, when considering the clinical picture of cholesterol granuloma in 1929 described it purely symptomatically as *blue drum* H V Forster in 1947 spoke of the black mastoid on the basis of the macroscopic appearance at operation In 1954 R R Simpson coined the term black cholesteatoma for cases of black cellular mastoid and suggested that all types of cholesteatoma may arise from a cholesterol granuloma In 1956 J F Birrell in association with G Young produced a terminological variation from black cholesteatoma to black cellular cholesteatosis Seventy-seven of J F Birrell's 11 cases had a typical cholesterol granuloma according to the histological evidence and the three illustrations In none of those 11 cases was there any evidence of cholesteatomatous matrix R R Simpson s suggestion that cholesterol granuloma is a precursor to cholesteatoma is possibly based on the following definition of cholesteatoma of the middle ear by A Eggston and D Wolff 'The lesion as found in the ear may be defined as an amorphous mass of desquamated epithelium, *cholesterin crystals* and wax, surrounded by layers of more recently grown stratified squamous epithelium, the whole forming an invasive cast of the cavity in which it lies." According to very accurate chemical experiments performed by A. Fazi of Pisa, B. Simonetta (1949) was able to show the term cholesteatoma to be a misleading one. "In two cases of large cholesteatomas the cholesteatomatous masses in their entirety were subjected to chemical analysis and this latter in fact revealed the presence of cholesterol, but in such minute traces as to be negligible." If cholesteatomas were to evolve from cholesterol granulomas then the metaplastic transition would have to include the dissolution and absorption of the abundant cholesterol crystals present in the granuloma. The ability of the human organism to destroy sterols appears, however, very doubtful. In the opinion of S. J. Thannhauser, one of the greatest authorities of the cellular lipoid metabolism, "destruction of the sterol skeleton in the intermediary metabolism cannot be accepted as a fact until the enzymes which should be involved in the process are discovered". R. R. Simpson's suggested transition from a cholesterol granuloma into a cholesteatoma thus seems less likely on these grounds.

There are at least two varieties of cholesterol granuloma, namely the genuine cholesterol granuloma and that occurring in association with a chronic suppurative otitis media.

G. E. Shambaugh first described the rare genuine cholesterol granuloma with the following clinical characteristics. A gradual onset of a conduction deafness without any signs of inflammation, a blue-grey discoloration of the intact tympanic membrane and radiological evidence of clouding of the more or less pneumatized mastoid. These changes are caused by an accumulation of brown-black slime and the formation of very vascular granulation tissue (Fig. 7) in the middle ear and its cellular extension. The granulation tissue contains numerous cholesterol crystals and giant cells. The bony walls of the middle ear are as a rule not affected.

Six cases of genuine cholesterol granuloma were seen and operated on at the Zürich clinic in the last six years. The above mentioned granulation tissue, rich in cholesterol, was found in every case and verified histologically. In none of our genuine cases was there any histological evidence of a cholesteatoma arising or even a mucosal metaplasia which might give rise to a cholesteatomatous matrix as a result of the presence of the granuloma. During the six-year follow-up period none of the cases showed any signs of a subsequent cholesteatoma formation. Any ætiological relationship between the rare genuine cholesterol granuloma and the common cholesteatoma is, therefore, considered most unlikely.

The second type of so-called associated cholesterol granuloma is much commoner. B. Simonetta (1932) has described a large cholesterol pseudocyst formed in a radical mastoid cavity, some time after complete healing of the wound. Such smaller cholesterol granulomas not infrequently occur in chronic middle-ear infections with or without cholesteatoma. They are usually distal to the main lesion in cells which have become shut off during incomplete sclerosis of the mastoid These cells, which no longer communicate with the middle ear, often contain sticky brownish sludge on a thickened and partially granulating mucosa The granulation tissue is again rich in blood vessels, cholesterol crystals and giant cells In cases where cholesteatoma and cholesterol granuloma co-exist, the division between the two pathological processes is sharp and striking, both structurally and in the distribution of cholesterol

This is illustrated by the case of a 23 year old man with a cholesteatoma in the right middle ear (Fig 8).

In spite of the proximity of the granuloma and cholesteatoma there is no evidence in these cases of epithelial metaplasia or of any metamorphosis of the granuloma into a cholesteatomatous matrix We agree with B Simonetta's statement "that in such a case the two manifestations, cholesteatomatous and cholesterinic, co exist, but remain separate, if they fuse at all the fusion is purely secondary when a cholesteatoma in its progression has eroded the bony walls of periantral cells which contained a cholesterol granuloma".

Thus biochemical, histological and clinical evidence all point against the possibility of cholesteatomas arising from cholesterol granulomas As the ætiology of the latter still remains to be elucidated we would suggest that the histologically descriptive term of *cholesterol granuloma* of the middle ear be used rather than "black cholesteatoma" or "black cellular cholesteatosis"

In spite of the evidence presented in favour of the immigation theory enthusiastic supporters of the metaplasia school could object that all the histological material showed only proliferation of the basal cells in Shrapnell's membrane as a result of inflammation Histological proof of the origin and development of cholesteatoma behind the intact drum is still lacking in man Such evidence may be available in animal experiments

Typical cholesteatomas in animals have been produced by the application to the external auditory meatus of cold or hot tar, benzpyrene, croton oil or olive oil or by injection of these substances into the middle ear (J Berberich (1927), H Hoshiya (1935), T Ohta (1940), R Schroer (1957)) The skin of the deep meatus proliferates in these experiments and grows into the middle ear through a myringotomy opening or spontaneous perforation No signs of metaplasia of the mucosa of the middle ear were found on any occasion In this connection a special mention must be made of the brilliant experiments performed by J Friedmann, who infected the middle ear in guinea-pigs with *Pseudomonas pyocyanea* and with *Streptococcus pneumonicus* He then found that "the stratified squamous epithelium from the external auditory meatus or from the tympanic membrane immigrated through the perforated tympanic

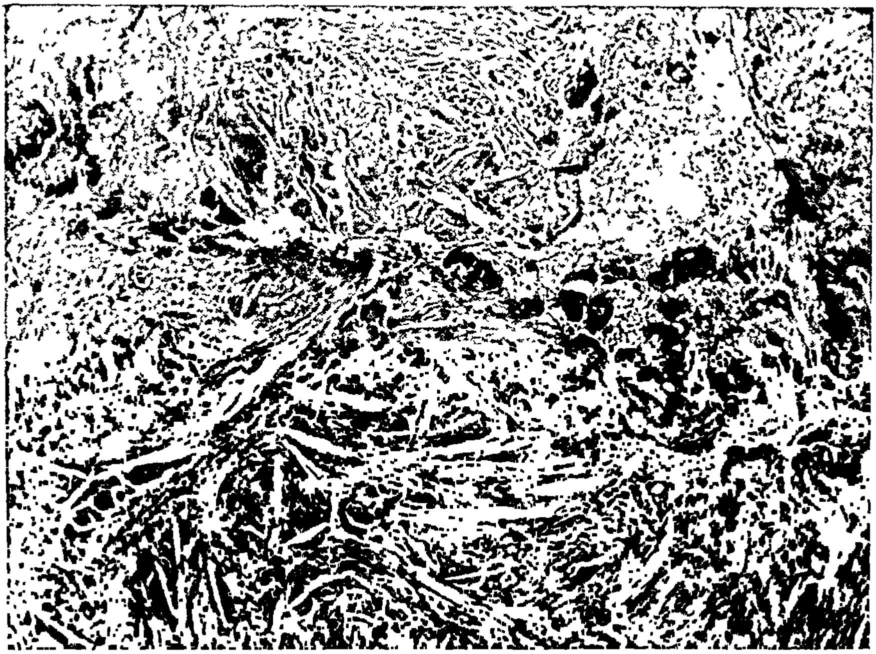

FIG. 7.
Genuine cholesterol-granuloma in the case of a 21 year old girl.

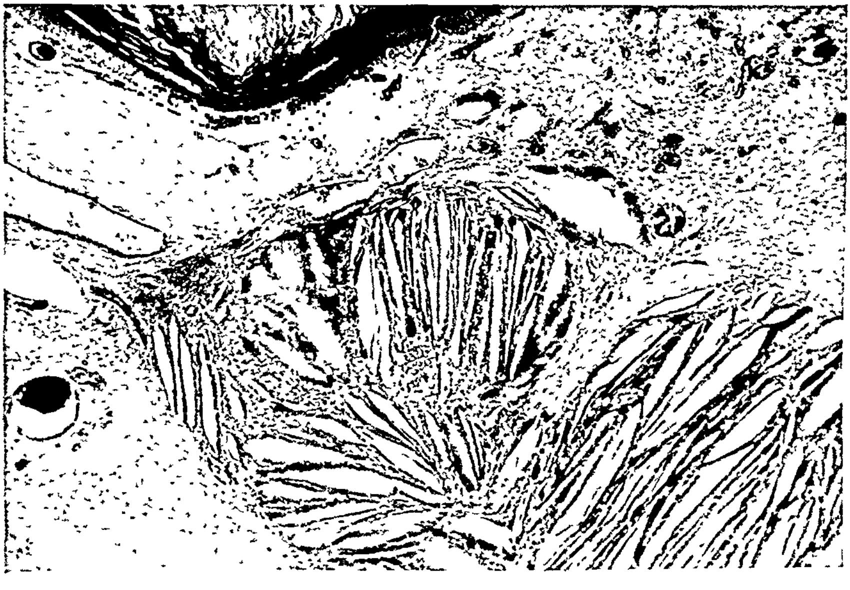

FIG. 8
Otitis media chronica cholesteatomatosa in the case of a 23 year old man. Associated cholesterol-granuloma in the neighbourhood of the cholesteatomatous matrix.

membrane into the infected bulla of the guinea-pig, reproducing here a typical aural cholesteatoma". J. Friedmann's experimental findings fit in very well with J. Habermann's classical observations in necrotizing otitis media of infants. Review of the literature shows that a tympanic perforation was present in all the experimental cases of cholesteatoma. The cholesteatoma developing behind an intact tympanic membrane, which is of special interest to us, has not yet been reproduced experimentally.

In order to test this possibility, we have opened the aural bulla in guinea-pigs and stuck a mixture of talc and fibrin in several places onto the internal surface of the intact tympanic membrane. This caused a mild foreign body reaction and in several animals granulation tissue developed between the drum and the internal wall of the middle ear (Fig. 9). After 15-20 days there occurs in some animals an active ingrowing of the epidermis from the intact tympanic membrane into the newly formed granulation tissue. The invading columns of basal cells divide into branches (Fig. 10) and the desquamated stratified squamous epithelium forms typical cholesteatomatous masses. Thus a mild inflammatory reaction resulting from chronic irritation may produce a cholesteatoma behind an intact tympanic membrane in the guinea-pig. Further details will be given in a special paper. There is no doubt that this experimental cholesteatoma results from an immigration of the squamous epithelium of the tympanic membrane stimulated by an inflammatory reaction. This makes the proposition that human aural cholesteatomas behave in much the same way all the more likely. The initial and the final stages of immigration of stratified squamous epithelium have already been determined in man. However, the intermediate stage of cholesteatoma formation behind the still intact tympanic membrane, which we have been able to demonstrate in experimental animals, has not yet received histological confirmation in man. In common with J. Friedmann we have not been able to find any evidence of mucosal metaplasia or cholesterol-granuloma in any of our guinea-pigs.

Clinical and histological study of normal and pathological human ears, supported by animal experiments, leads to the conclusion that as a rule all types of cholesteatoma of the middle ear develop by immigration of stratified squamous epithelium from the epidermis of the external auditory meatus or the tympanic membrane. Within the middle-ear cavities the active growth of the matrix is enhanced by submucous connective tissue filling the incompletely pneumatized attic and epitympanic cells. As seen in the guinea-pig, the cholesteatomatous matrix may also continue to grow within newly formed granulation tissue.

The practical application of the immigation theory to the elimination of the risks of cholesteatomas, achieving a dry ear and improving the hearing must now be considered.

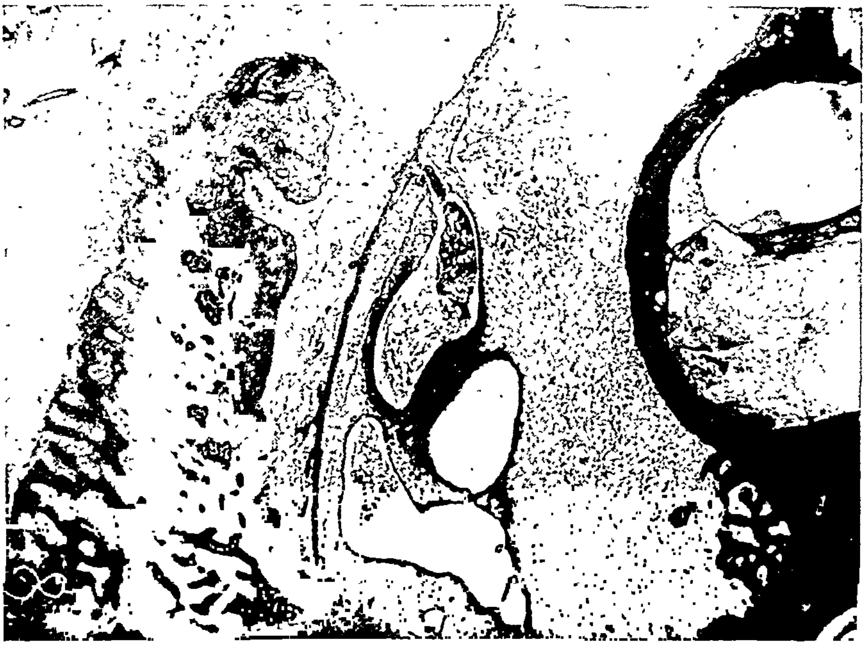

FIG. 9.
Vertical section through the middle ear of a guinea-pig. The external meatus is narrowed to a slit but the tympanic membrane, is intact. Active ingrowing of the epidermis from the intact tympanic membrane into the newly formed granulation tissue.

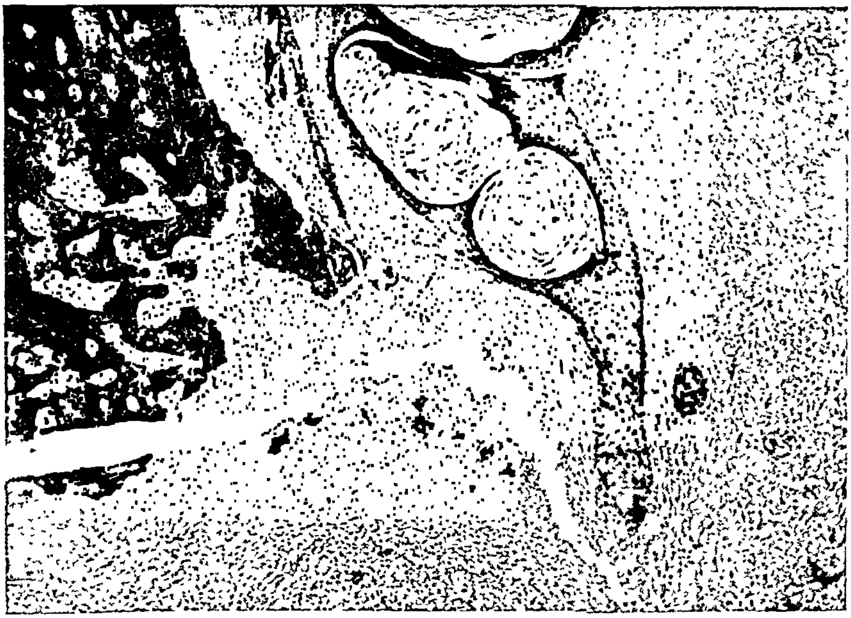

FIG. 10.
Higher magnification of Fig. 9 (vertical section through the middle ear of a guinea-pig). Experimental cholesteatoma develops behind the intact tympanic membrane.

The dangers of cholesteatoma depend on the growth of the matrix into the middle ear and its further multiplication there. In order to eliminate this dangerous process it is essential thoroughly to eradicate the matrix, the submucosal connective tissue which facilitates its spread, and all the granulation tissue in the surrounding cells. Complete removal of the cholesteatoma can only be achieved surgically. As cholesteatomas usually begin in childhood, the sooner they are removed the less extensive will be the resultant damage to the ear. The most important therapeutic principle is, therefore, *all forms of cholesteatoma should be treated surgically as early as possible.*

The cholesteatomatous matrix grows in depth, affecting all the cell groups still available in the mastoid process of the child. In order to eradicate the disease all the cells must be meticulously opened and cleared out. This important step in the operation is often technically difficult, especially in the sub- and retrofacial and the hypotympanic peritubal cells. We attempt to achieve the last two aims of treatment, namely the establishing of a dry ear and improving the hearing by means of tympanoplasty. In earlier years to ensure a dry ear, many methods of shutting off the Eustachian tube through plombage of its tympanic opening were tried, often without success. Today the newly fashioned cavum tympani is shut off more easily by means of a new tympanic membrane. This new membrane fashioned from a skin flap of the anterior meatal wall bridges the hypotympanum and is inserted along the external semicircular canal. The membrane covers the facial ridge, and passing forwards and upwards it will lie somewhat lateral to the original tympanic annulus on the anterior meatal wall.

Improvement in hearing depends on the patency of the tube, integrity of the ossicular chain and the function of both the inner-ear windows. A stenosed Eustachian tube is dilated with silver sounds and it is held open by passing a small acryllic tube down and out through the nose. In order to keep the newly fashioned cavum tympani in communication with the round window, it is inflated with air through the indwelling acryllic tube for the first ten days. Unfortunately the ossicular chain is very often interrupted by the cholesteatoma, especially at the point where the long process of the incus articulates with the stapes. Thus only the stapes or sometimes only the mobile footplate remain available for the conduction of sound. Accordingly the new tympanic membrane flap is either placed directly onto the stapes or connected to the footplate by the interposition of a columella. Finally the niche of the round window, which is frequently overgrown, must be freed if an improvement in the hearing is to be achieved. Further technical details of the tympanoplasty are then dealt with as they arise in the course of the operation.

Fig. 11 gives the surgical results which have been achieved in 88 cases of aural cholesteatoma.

Number of cases	88 =	100 per cent.
(1) Endocranial danger definitely eliminated	88 =	100 per cent.
(2) Ear dry	78 =	88·6 per cent.
(3) Tympanoplasty successful	69 =	78·4 per cent.
(4) Hearing improved	44	50·0 per cent.
(*a*) More than 20 db.	10 =	11·4 per cent.
(*b*) 10-20 db.	19 =	21·6 per cent.
(*c*) Until 10 db.	15 =	17·0 per cent.
(5) Hearing unchanged	25 =	28·4 per cent.
(6) Hearing worse	19 =	21·6 per cent.
(*a*) Until 10 db.	11 =	12·5 per cent.
(*b*) More than 10 db.	8 =	9·1 per cent.

FIG. 11.

Better ear surgeons will get better results.

Summary

Cholesteatomas in the middle ear are classified in a larger, less dangerous group with large upper marginal perforations and in a smaller more dangerous group with small upper anterior or posterior perforations.

Clinical and histological studies have shown that as a rule cholesteatomas with large and with small upper marginal perforations develop by immigration of stratified squamous epithelium from the external auditory meatus or tympanic membrane. This process takes place in some cases during acute necrotizing otitis media through a large upper marginal tympanic perforation. Aural cholesteatomas with small upper marginal perforations develop insidiously as a result of recurrent acute catarrhal inflammations of the middle ear in childhood. The basal cells of the intact pars flaccida of the ear drum multiply and grow into the submucous connective tissue filling the incompletely pneumatized attic, or into newly formed granulation tissue, where they continue to grow into a cholesteatoma. These cholesteatomas, which have primarily originated behind an intact tympanic membrane, usually break through into the external auditory meatus and give rise to a small, upper marginal perforation. The formation of cholesteatomas behind an intact tympanic membrane has been reproduced experimentally in guinea-pigs by setting up a mild inflammatory reaction in the middle ear.

The risk of intracranial complications is ever present in all types of cholesteatoma because of the invasive tendency of the matrix. In order to eliminate this risk and to arrest the persistent otorrhoea, early surgery is indicated. Tympanoplasty offers the best means of improving or at least preserving the hearing, depending on the integrity of the ossicular chain, the state of the round window, and the patency of the Eustachian tube.

The operative results obtained at the Zürich E.N.T. Clinic during the last two years are presented.

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ATTIC CHOLESTEATOSIS

By A. TUMARKIN (Liverpool) *

I AM very honoured to be taking part in this symposium and I feel it is a particular privilege to follow Professor Rüedi who has taught us all so much about the pathology of the mastoid bone. I have been an earnest student of his work ever since 1937 when he published his famous monograph on the development of the middle-ear spaces (Rüedi, 1937).

I have to admit that my own views on the nature of cholesteatosis have altered very considerably during the past few years, I have been greatly influenced by Professor Rüedi and even more so by Dr. Friedmann (Friedmann, 1955, 1957), who has converted me to the immigration theory (Fig. 1). Unfortunately the problem is complicated by the fact that the term cholesteatosis, as used to-day, covers two entirely different entities.

FIG. 1.
Cholesteatosis includes two different entities

EPIDERMOSIS	CHOLESTEROL GRANULOMA
True epidermis derived from meatal skin by *immigration*	(Cholesterol crystals, granulations, giant cells, *blood pigments*)

In the first the attic is filled with epidermis derived from the skin of the meatus by a process of immigration. For clarity I shall refer to this as epidermosis. Secondly there is cholesterol granuloma characterized by cholesterol crystals, granulations, giant cells and blood pigments. I have been studying this condition for the past 10 years. Fig. 2, is the slide I showed on this screen eight years ago. At that time I said "This picture differs markedly from cholesteatosis" (Tumarkin, 1950). Unfortunately many writers, being impressed by the cholesterin crystals do regard it as a variety of cholesteatosis, and this leads to ambiguity. To avoid confusion, therefore, I shall avoid the word "cholesteatosis" wherever possible. I shall speak instead of "epidermosis" or of "cholesterol granuloma" as the case may be (Fig. 3). Any theory to be complete must explain them both; it must explain why epidermosis is so insidious and why it is usually, although not invariably, associated with a hypocellular mastoid.

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* Read at the Section of Otology, Royal Society of Medicine, 7th February 1958.

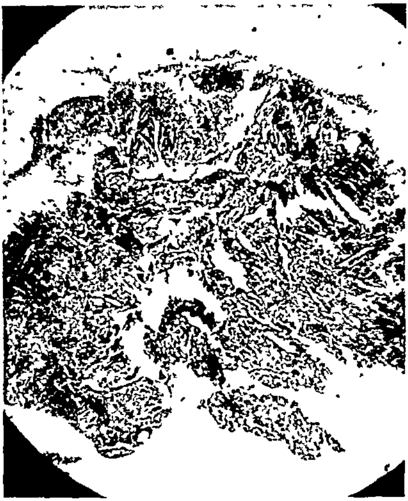


FIG 2

Micro-photograph of granulations removed from a case of chronic enclosed mastoiditis The disease had been present for over a year and yet the drum was intact Diagnosis was based on the lack-lustre gun metal appearance of the drum plus typical clouding of a pneumatized mastoid

FIG 3

A Complete theory of Cholesteatosis must explain

(1) Epidermosis
(2) Cholesterol granuloma
(3) Insidious onset
(4) Hypocellularity
(5) Marginal perforation
(6) Drum retraction

It must explain why the perforation is characteristically marginal and why the drum is so frequently retracted

Finally it must take note of the fact that dry retraction of the drum head—including, of course, dry perforation of Shrapnell's membrane—is by far the commonest pathological condition encountered within the middle ear For example, in unilateral epidermosis we usually find that the opposite ear shows a retracted drum

I suggest that dry collapse is the essential primary pathology and epidermosis only occurs when some extra secondary factor is superadded

Our first task, therefore, is to explain "dry collapse" and then to consider what converts this into active epidermosis.

I take my stand on the Bezold Habermann theory which postulates obstruction in the eustachian tube with secondary intratympanic vacuum, a condition which is extensively common in children, especially slum children who suffer from nasopharyngitis and adenoid hyperplasia. When the eustachian tube is chronically obstructed the drum tends to cave in, either at the pars tensa or at the pars flaccida. At the same time the process of pneumatization falters and may entirely cease.

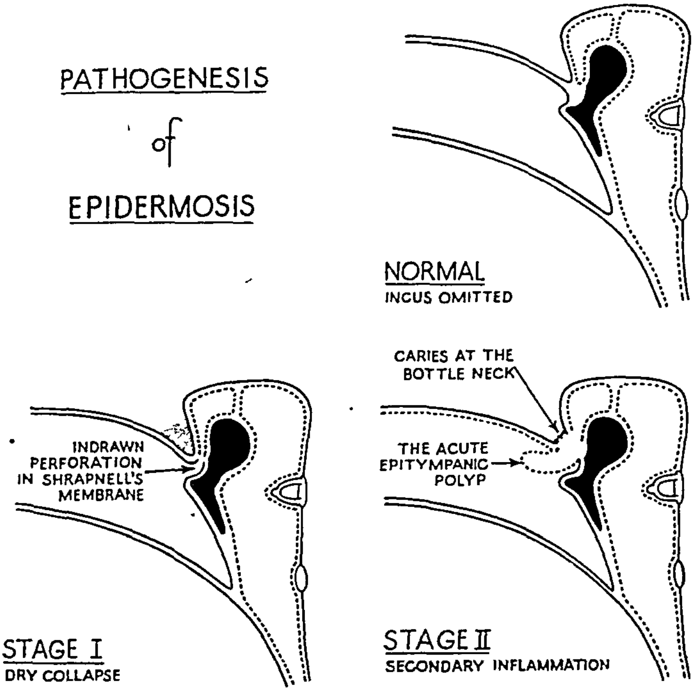

PATHOGENESIS
of
EPIDERMOSIS
NORMAL
INCUS OMITTED
INDRAWN PERFORATION IN SHRAPNELL'S MEMBRANE
STAGE I
DRY COLLAPSE
CARIES AT THE BOTTLE NECK
THE ACUTE EPITYMPANIC POLYP
STAGE II
SECONDARY INFLAMMATION

FIG. 4.

The idea that a vacuum—by its simple mechanical effect could inhibit pneumatization is resisted by many authorities. I myself find it entirely plausible and recently Ojala (1957) has proved conclusively that pneumatization in the humerus of a growing chicken can be inhibited by simply blocking the air channel to the developing air cell system. He concluded that exactly the same thing can occur in the mastoid of a growing child.

In many cases nothing more happens and the individual may go through life unconscious that his mastoid is hypocellular, his drum indrawn and his hearing slightly impaired (Fig. 4).

He is, however, vulnerable—especially if the collapse has gone on to actual perforation of the drum head. Suppose he succumbs to infection at a later date. Inflammatory exudate forms within the middle-ear cleft and leaks away through the preformed perforation. Thus there is no distension of the drum, no otalgia. Instead there is the typical insidious otorrhea followed by the sprouting of granulations from the carious bone surrounding the bottle neck of the perforation. But the bottle neck is already lined by the epidermis which has been sucked in by the preceding vacuum. Inevitably that epidermis responds in accordance with its nature—it tries to grow over and protect raw granulations—it enters the attic and epidermosis begins. Thus the pathogenesis of epidermosis requires a preliminary stage of dry collapse and later an inflammatory process to set it alight. Some authorities maintain that acute inflammation can produce epidermosis de novo—that is without any such preliminary dry collapse. Dr. Friedmann has produced typical epidermosis in the auditory bullae of healthy guinea-pigs by the artificial introduction of pyogenic organisms and Professor Rüedi believes that a violent necrotizing tympano mastoiditis can destroy the membrane right down to the annulus and so permit meatal skin to invade the middle ear. I think this must be uncommon. Acute suppurative otitis media tends to burst the pars tensa at the centre. Epidermosis cannot occur unless the resulting central perforation spreads right out to the annulus. Such a disastrous spread is unlikely in this era of antibiotics, and any epidermosis which did occur would bear little resemblance to the familiar attic epidermosis which we are now discussing.

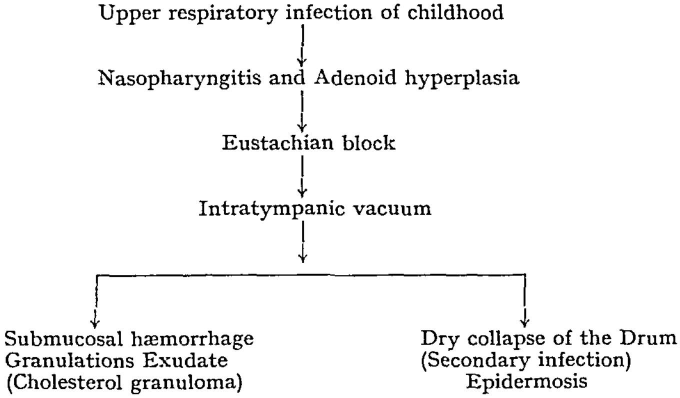
Apart from acute suppuration, however, there are other exudative reactions that can contribute to the development of cholesteatosis. For example in response to intratympanic vacuum the mucous membrane must become engorged and in some cases petechial hæmorrhages will occur. Small granulations will spring up and the whole cleft may become filled with sanious exudate. This possibly explains why blood pigments figure so prominently in cholesterol granuloma. It may explain the curious condition of the blue drum in which the mastoid is more or less filled with an indolent sanious exudate. Fig. 2 was obtained from such a case. The patient had been under observation for over a year during which time there has never been any otorrhœa whatsoever. When we opened the mastoid we found typical cholesterol granuloma. This sort of sterile mechanical process is comparatively rare. More commonly, possibly as a response to virus infection, the middle ear becomes filled with a glue-like exudate. Within recent years we have been studying this condition of exudative otitis media amongst the children of the Liverpool slums and have been appalled at its frequency. Treatment is difficult and protracted, nevertheless it can be extremely rewarding. Unfortunately in some cases the exudate recurs obstinately and finally symptoms of a more violent nature appear. The child begins to suffer with recurring bouts of otalgia and finally otorrhœa. We have explored the mastoid in some cases and have found all types of pathology from the acute purulent granulations to the characteristic granuloma. We have, therefore, come to regard cholesterol granuloma as merely one aspect of a complex pathological process. We believe, however, that it occurs in its purest form when inflammation is minimal, more especially when a cellular mastoid bleeds under the mechanical effect of a vacuum.

Lastly this vexed question of pneumatization. I believe the hypocellular mastoid is always abnormal. It results either from inhibition of pneumatization, as I have just described, or from condensing osteitis following inflammation. And so it is not surprising that cholesteatosis is commonly associated with a hypocellular mastoid. Nevertheless that association is not invariable. Why should it be? The pneumatic system develops quite rapidly and if a child reaches the age of 5 or 6 before developing eustachian obstruction it will have acquired a comparatively large pneumatic system.

Thus we need not be surprised to find epidermosis within a fully cellular mastoid and, since the delicate trabeculae of the pneumatized bone can offer so little resistance to the spread of disease, it is in these very cases that we find the most widespread invasion of the whole petromastoid structure.

These, in my opinion, are the basic factors responsible for cholesteatosis and they enable us to answer the question as to the relation between epidermosis and cholesterol granuloma. We see that from one point of view they are different insofar as epidermosis arises by immigration of meatal epithelium, following dry collapse, whereas cholesterol granuloma is a variety of exudative mastoiditis (Fig. 5). On the other hand they are closely related since both are produced in the growing child's mastoid by

FIG. 5.
Pathogenesis of Cholesteatosis.



the same basic disease Thus in listing the aural complications of the upper respiratory disease of childhood, although we demarcate many different entities from dry collapse to acute suppuration we should not regard our demarcations as water tight We are dealing with a continuous spectrum of disease and we must not be surprised to find elements of more than one type present in any given case

MY POINT IS THAT EVERY ONE OF THESE CONDITIONS IS A RESULT—WHETHER IMMEDIATE OR REMOTE—OF THE DIFFUSE UPPER RESPIRATORY DISEASE OF CHILDHOOD

I now turn to the question of management

Thirty years ago this discussion would have been dominated by the intra cranial complications That I make no further reference to them is a measure of the great progress that has been made in Otology (Fig 6)

FIG 6
Types of Attic Suppuration

(6) Advanced disease with intracranial complications
(5) Advanced disease without intracranial complication but with necrotic ossicles ulceration of pars tensa as well as pars flaccida
(4) Advanced disease with necrotic ossicles perforation pars flaccida but intact pars tensa
(3) Early disease with viable ossicles and intact pars tensa
(2) Combined attic epidermosis and mastoid Cholesterol Granuloma
(1) Acute Epitympanic polyp

Nevertheless we still encounter far too many cases in Group V, i e advanced pathology with disorganized ossicles and perforations in the pars tensa and the pars flaccida In the past we have been happy enough to secure a dry ear in such cases, believing that sacrifice of the ossicular chain inevitably meant a poor functional result Nowadays however, hopes have been raised high by the surgical triumphs of our Continental friends It seems, however, that the astounding successes that can undoubtedly be achieved with other types of pathology are not so easily obtained in epidermosis The basic difficulty in reconstructing a middle ear invaded by epidermosis is the need to eradicate completely all traces of the invading epidermis before applying a skin graft Zollner himself spoke very feelingly on this point at the recent International Congress

> "Wherever Cholesteatoma can be removed radically primary plastic surgery is promising—but if the hypotympanum must be curetted radically we often encountered recurrences of Cholesteatoma underneath the plastic flap, despite the most careful curettage We, therefore avoid any inclusion of the skin within the area of plastic surgery
>
> "Our procedure has become more and more conservative as the years have gone by, but this consideration must not prevail at the expense of the dependable healing of the disease process That is why we do not cover a recessus epitympanicus, affected with Cholesteatoma, with a plastic flap F Zollner Int Cong 1957

Recently Richtner (1957) published a small series of cases dividing them into seven groups. He used mainly Type IV Tympanoplasty of which he speaks very highly. Nevertheless he admits that his worst results were achieved in cholesteatosis, only one case in seven improved.

My own experience of these operations is negligible and I look forward to hearing from others who can speak with authority on this important problem.

Suppose now we are dealing with stage IV in which the pars tensa is intact, but the ossicles are not viable. The incus and head of malleus must be removed and the question arises—shall we carry out Type III Tympanoplasty? Here again the problem of completely eradicating the epidermis confronts us—but in addition the use of a free graft under these circumstances is open to criticism.

This graft serves three purposes:

(1) It replaces defects in the pars tensa.
(2) It epithelializes the mastoid cavity.
(3) It provides a mechanical bridge between the reconstituted pars tensa and stapes.

But in these cases the pars tensa is intact. There can surely be no gain in first denuding the drum of its epithelium and then skin grafting it?

Again, the survival of the free graft in the mastoid cavity is, to say the least, precarious. I believe that the tympanomeatal flap—as in fenestration—has a much better chance of survival, and the same argument applies to the third function of supplying a mechanical bridge. Thus, from every point of view, a pedicle flap should be superior to a free graft, whenever the pars tensa is intact.

The fashioning of such a flap is complicated by the presence of a perforation in Shrapnell's membrane and the tissues are more friable than normal. Nevertheless the flap can be made and in more successful cases the perforation should heal over. This is the sort of tympanoplasty I am trying at present and I am sure that many others are doing likewise.

Next, suppose we are dealing with Stage III in which the pars tensa is intact and the ossicles are viable. In the past I have aimed at exposing the invading epidermis wherever it ramifies. My idea was simply to evacuate all debris and to provide access to the skin lined cavity, so that suitable toilet could prevent the reaccumulation of squamae and ultimately the ear could heal completely. It was with this idea that I proposed the conservative permeatal atticotomy (Tumarkin, 1947). There was nothing new in that idea of exposure and conservation. It has been pursued by many surgeons both before and since my time. Stacke himself attempted something of the sort. Dundas Grant alway advised preservation of the matrix and indeed that idea is implicit in the various conservative radical operations that have been proposed from time to time. Dingley (1955) recently claimed excellent results from this sort of conservative surgery and Baron (1949) of San Francisco ardently supports this view. This sort of operation can frequently give highly satisfactory results Unfortunately the exposed ossicles are vulnerable and demand periodic toilet I have been saddened to see, on more than one occasion, an apparently brilliant result return years later with a neglected cavity full of stinking debris I have learnt to choose my cases for this sort of operation, not only on their pathology, but also on their social status and personal hygiene (Fig 7) I have tried to improve matters by covering the ossicles with a tympano-meatal flap I call this segregation of the ossicles and I described it to the

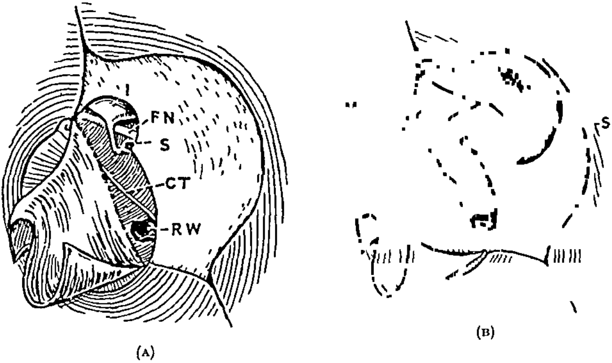


Fig 7

(A) The flap has been elevated down to the annulus and turned forward across the drum bringing into view the posterior third of the tympanic cavity The first semilune has been removed bringing into view the long process of the incus I and the facial nerve F N
R W —Round Window C T —Chorda Tympani S —Stapes

(B) The attico antrotomy has been completed The incus is fully exposed lying on the semi circular canal S C The head of the malleus M is visible Diseased cells are removed wherever they ramify and finally the flap is turned into place over the ossicles so as to segregate them from the mastoid cavity

Society here eight years ago. You will observe the similarity to a tympanoplasty Type II except, of course, that a sliding graft is used instead of a free graft The results of this operation were, on the whole, disappointing For one thing, at that time I was working by a purely permeatal approach through aural specula Such an approach does not permit the microscopic survey which is so essential to reconstructive surgery of the ear I have now abandoned the permeatal route and hope to do better with a wider approach of more orthodox incisions Even so, however, I am dubious of the value of applying grafts to the ossicles under such circumstances Not only is there risk of burying epidermic islands, but there is a real risk that the whole ossicular mass will become buried and bound down by adhesions. If so we are on the horns of dilemma. If we expose the ossicles they are vulnerable, if we graft them they solidify.

In the face of this dilemma it is natural to enquire whether it might not be possible in some way to retain the outer attic wall, so as to protect the ossicles from the external world. In the past I have always assumed that this was out of the question.

Consider the patient who presents with an acute epitympanic polyp, giving a history of quite recent insidious otorrhœa. I used to approach this sort of case with pessimism. Despite the short history I always felt that there was inevitably so much deep pathology within the mastoid that conservative measures must be futile. And yet everyone knows that such cases can sometimes be cured by quite simple means. Since my conversion to the immigration theory I have realized why this must be so. The mastoid may be hypocellular and it may be inflamed. Nevertheless the major pathology, i.e. the epidermic invasion, may have only just begun at the surface and, if that is the case, it can be halted and repulsed. With this altered view point I have been attacking these cases with renewed vigour. I flood them with antibiotics and then, under local anæsthesia, with adequate magnification, I remove the epithelium from the margins of the perforation, suck out the presenting polyp, and curette the bony margins. Providing the after care is intensive and meticulous this simple procedure can yield most gratifying results and I commend it to your attention.

In all these groups the dominating pathological feature is the epidermosis and I have made no reference to the state of the mastoid itself. In Group II, however, in addition to the attic epidermosis there is cholesterol granuloma in a cellular mastoid. This is particularly liable to occur in children and is one of the most interesting and important groups. Mr. Birrell has made a special survey of these children and, like everyone else, I am looking forward to hearing his account of his work and so I make no further reference to this group.

How then do we manage cholesteatosis?

In Group VI we are desperately fighting for the patient's life with little thought indeed for his hearing.

In Groups V and IV all the resources of modern microsurgical technique must be deployed and, even so, the functional results leave much to be desired.

In Group III satisfactory results can be obtained, but only at the cost of subsequent perpetual vigilance and even when the pathology is minimal, as in Group I the healed ear is still in a precarious condition. Is this the best we can hope for? Surely not—If what I have said is correct cholesteatosis takes its place along with chronic tonsillitis and chronic sinusitis and, indeed, bronchiectasis, as one of the complications of the upper respiratory disease of childhood Surgery—whether it be tonsillectomy, mastoidectomy, pneumonectomy—however flamboyant is a confession of defeat It can indeed be harmful if it diverts our energies from the humble drudgery of conservative and preventive treatment

Our work in the Liverpool slums has convinced us that cholesteatosis is preventable Our aim at present is to cure the underlying diffuse disease if possible without recourse to surgery This we can often achieve Nevertheless there are still many cases in which despite our best efforts, the disease progresses implacably It is obvious that we have much more to learn about its pathogenesis Perhaps most urgent of all is our need to elucidate the role of the viruses Some day we shall solve these problems Some day it will be possible to immunize children against this respiratory disease

When that day dawns I believe that it will mean the end of chronic ear disease and that, in my opinion, is the only satisfactory way to manage cholesteatosis

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CHOLESTEATOSIS OF THE ATTIC

By J. F. BIRRELL (Edinburgh) *

EXPERIENCE with mastoid surgery in children has shown me that in approximately one half of the cases in whom true squamous cholesteatosis has been present in the attic, the mastoid has been pneumatized, often extensively so, and my remarks will be confined to such cellular cases. I was interested to read that Professor Rüedi (1957) found, in a series of 32 children between the ages of 5 and 10, who required radical surgery, that 55 per cent. were associated with cellular mastoid processes.

The extensive use of antibiotics, usually in children oral antibiotics, for acute otitis media, has greatly reduced the number of acute mastoid operations which we have to perform, but the inadequate dosage which is all too frequently administered has led to an increasing number of cases of subacute enclosed mastoiditis. We see a great many children, with a history of recurring episodes of otitis media, whose drums never return completely to normal between attacks. During the acute phase the drum reddens and bulges in the usual way, but if treatment is stopped too early the membrane may never regain its normal lustre and contour, but may remain dull and indrawn, often acutely. This state of affairs is not invariably relieved by attention to the tonsils and adenoids, by washing out the antra, or by inflating the Eustachian tubes. The deafness which persists in association with the retracted membrane, has been treated in some centres by application of radium to the lymphoid tissue in the fossa of Rosenmuller.

Not infrequently an X-ray of the mastoids in such cases, taken in the quiescent phase, may show a pneumatized process with some blurring of the cells and destruction of the intercellular walls. At operation these findings are confirmed, and the infected cells are found to be of a yellowish-green colour containing some thick mucoid secretion which is usually sterile. These infected cells lie in tracks extending from the mastoid antrum to the tip and towards the petrosal angle. The mucosa of the mastoid antrum and attic is thickened, and shows on section the appearances of chronic inflammation. Granulations may be found, but white squamous cholesteatosis is never present.

Uncommonly a child with a similar history of recurring pain and deafness may show, in the quiescent stage, a blue drum which may be of normal configuration with a cone of light. X-rays will show blurring of the cells on the affected side, and at operation this blurred appearance is found

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* Read at the Section of Otology, Royal Society of Medicine, 7th February 1958.

to be due to the cells being filled with a black homogeneous substance which, as Ranger (1949) described, may easily be removed from the cell leaving an apparently healthy lining, but which, in my experience of some half-dozen of these cases, is often associated with a break-down of the intercellular walls in tracks similar to those already described. In these cases, there is a collection of sterile glairy secretion of a greenish-black colour, showing under the microscope superficial cells, squamous in type, and often nucleated. Similar fluid may be found at the aditus, aspiration through which will often clear the middle-ear cavity to a large extent of its collection of fluid. Again, the mucosa of the antrum and attic is thickened to the naked eye, but section of it will show the giant cells and cholesterin spaces of the condition known as cholesterin granuloma.

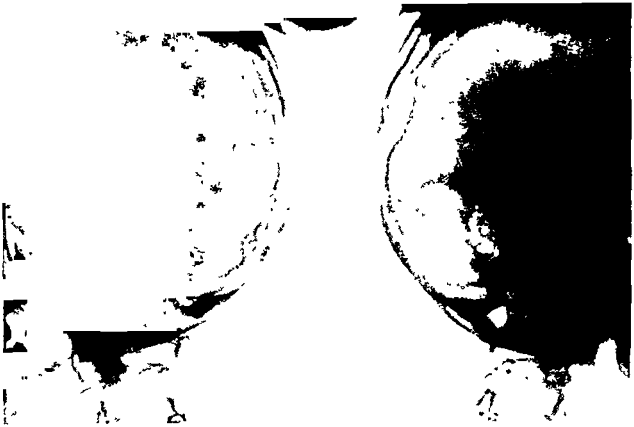

FIG 1

Radiographs of a child with a blue drum to show involvement in a cellular mastoid

Identical mastoid radiographs and operative findings are not infrequently met with in children with a similar history in whom the membrana flaccida remains slightly full, with small dilated vessels running across it, or in whom there is a perforation of Shrapnell's membrane, or of the postero-superior part of the membrana tensa, with a presenting granulation. Microscopically, in addition to the features of cholesterin granuloma in the black cells a partial squamous metaplasia of the thickened epithelium of antrum and attic may be found. Even when such a perforation exists true white squamous cholesteatosis need not be present in the attic.

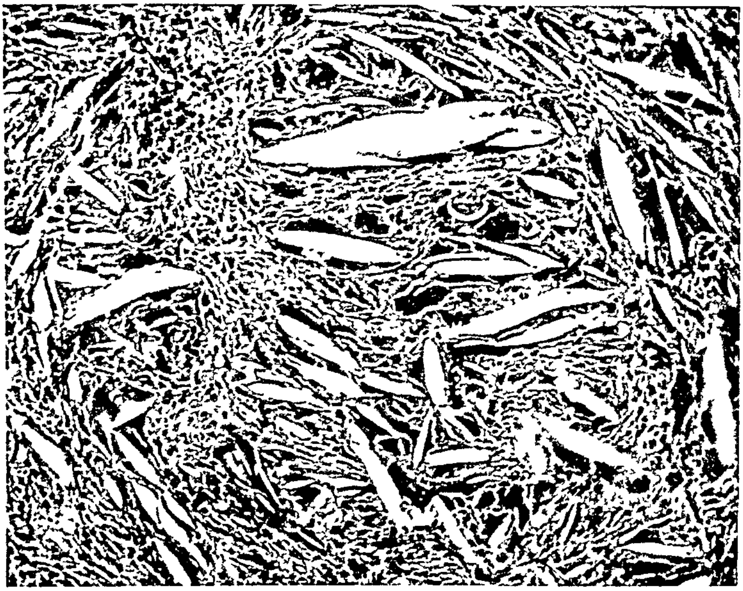

FIG. 2.
Cholesterin granuloma.

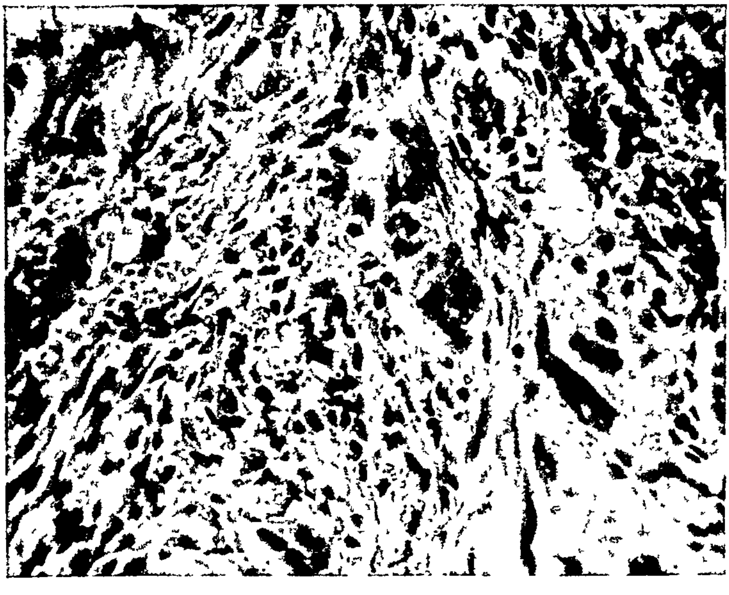

FIG 3
Cholesterin granuloma showing hæmosiderin.

On the other hand, such a perforation is much more commonly associated with true squamous cholesteatosis in the attic, and, in my experience, this is invariably accompanied, in those children with cellular mastoids, by the presence of black cellular necrosis more superficially in the mastoid process I have never found true white cholesteatosis in a child's cellular mastoid which has not been associated with cholesterin granuloma in black cells nearer the periphery, and especially in the tip and petrosal angles Sections from each area show appearances typical of each condition, coils of well-differentiated squamous epithelium in the attic, and giant cells, cholesterin clefts and often hæmosiderin in the mastoid cells

Once established in the attic, the squamous cholesteatosis spreads throughout the mastoid in the usual way, and undoubtedly, if the case were not operated upon it would, in time, complete Nature's mastoid This may happen in early childhood, and I recall one boy, aged 3, in whom the white mass had filled the entire mastoid with the exception of a layer of black cells immediately below the surface of the mastoid bone

It would seem to me that this series of conditions must be interrelated, and that, in cellular mastoids at least, it should be possible to trace the course of events All children with the history of recurring attacks of otitis media, especially if unilateral, or, indeed, of deafness in one or both ears, who do not respond to attention to the nasopharynx, pharynx and nasal sinuses, should have their mastoids X-rayed If the radiographs show healthy pneumatization, no mastoid operation is called for Should the films show bilateral acellular mastoids, a decision to *operate will depend on the history and the appearance of the drums in the* quiescent phase But if the mastoids are cellular, and if one or both should show evidence of blurring or of cellular destruction, exploration is called for, and the mastoid pathology at operation will justify this decision

There is little difference in the history, clinical examination and radiographic appearances between the subacute enclosed mastoiditis with chronic inflammatory changes, and that with cholesterin granuloma The main difference at operation is the obvious black discolouration of the cells in the latter condition due to the appearance of cholesterol crystals and hæmosiderin Ranger (1949) and Simpson (1954) have both speculated on the cause of these The blackish substance appears to be altered blood, but why does it arise? And why in one instance is it also found in the middle-ear cavity causing a blue drum, but in another it is limited to the subcortical cells while the attic contains white squamous cholesteatosis? I am informed that it is highly improbable that the mere presence of cholesterol in the secretion of the mucosa of the middle ear could predispose to hæmorrhage Further, as blood cholesterol shows wide normal variations, it appears to be unlikely that these children would exhibit abnormal blood cholesterol values

Again, the only difference between the black cellular case without

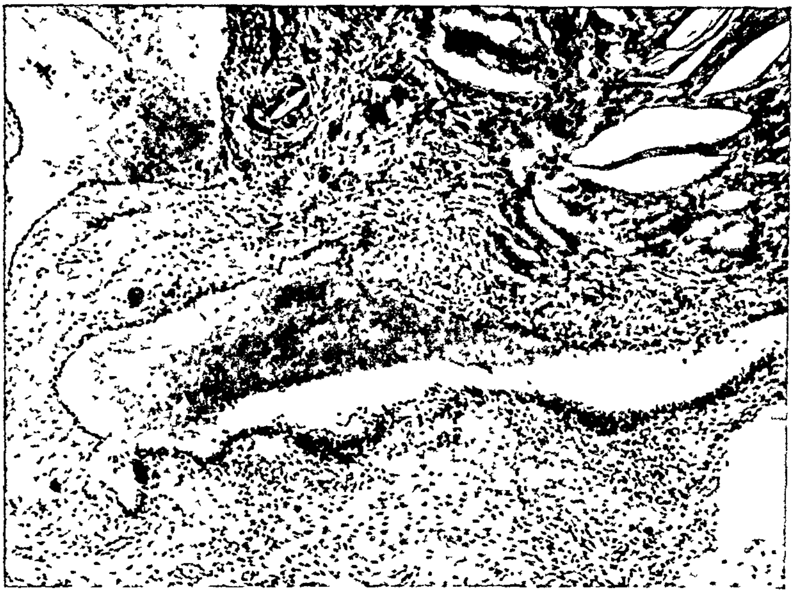

FIG 4
Partial squamous metaplasia in a cell, with cholesterin granuloma adjacent.

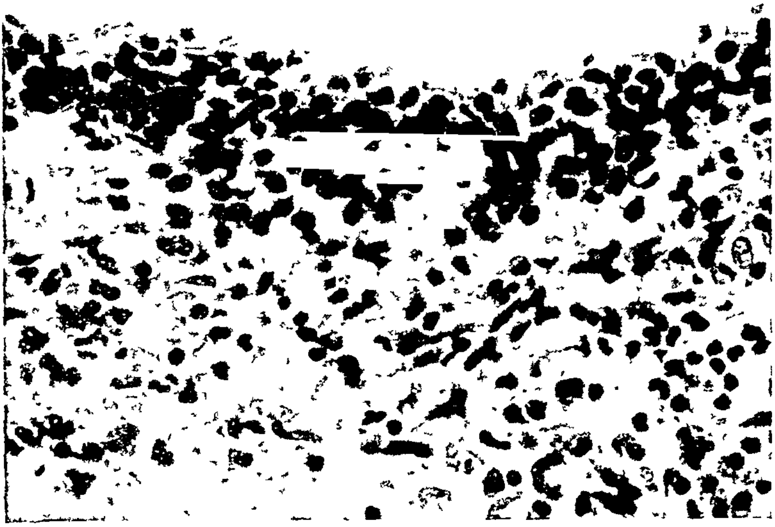

FIG. 5.
Squamous metaplasia Note the absence of true "prickle" cells

squamous cholesteatosis and one with, is that I have never yet found true white squamous cholesteatosis in association with an intact drum. Squamous metaplasia may be shown histologically in the mucosa of the attic and antrum, but this has failed to show prickle cells unless the drum is perforated and squamous cholesteatosis exists From this, in the present state of knowledge, one must deduce that, in cellular mastoids, a perforation in the postero-superior quadrant or in Shrapnell's membrane, is a necessary precursor to the development of squamous cholesteatosis Yet such a perforation is not the starting point of cholesterin granuloma, as this has been found in a number of mastoids without any history of discharge

Why does the drum perforate in these situations? The normal drainage of the mastoid cells is through the antrum, attic, middle ear and Eustachian tube, and it seems reasonable to suppose that this black secretion drains by the same route, seeping slowly through the antrum to lie in the attic Lodged there it will cause deafness, and possibly, in the presence of infection, pain It will give rise to a bulging of the membrana flaccida, and it may render this membrane so sodden that either it perforates spontaneously, or that a further upper respiratory infection results in its perforation Professor Ruedi (1957) has suggested that inflammation in the middle ear may initiate a proliferation of the basal cells within the epidermis adjoining the tympanic membrane, or within an intact Shrapnell's membrane, and that upon this cholesteatosis grows initially It may be that this secretion from cells showing histologically cholesterin granuloma will predispose to such a proliferation and eventual perforation through which an immigration of squamous epithelium may take place At the moment this seems more probable than that the squamous metaplasia within the attic and antrum proceeds to a well differentiated epithelium complete with prickle cells

In this series I have been greatly helped by the reports from our pathologist, Dr Agnes Macgregor, who has frequently commented on the absurdity of using the term cholesteatoma, or even cholesteatosis, for a condition in which cholesterin clefts are not, as Simonetta (1949) has shown, part of the typical histological picture On my suggestion we have adopted the term ' squamous epitheliosis" for the condition to which I have referred in this paper as ' true white squamous cholesteatosis' , as we believe that the term squamous epitheliosis more accurately describes the pathology without dogmatizing on the ætiology

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NASOPHARYNGEAL FIBROMA

WITH A REPORT OF THREE CASES

By JAMES P. FRASER (Glasgow)

NASOPHARYNGEAL fibroma is a condition which, according to most authorities, can be classed as rare in this country, and indeed, it has been claimed that it is becoming even more rare. Certainly, the bulk of the literature on this subject has come from abroad, and from the large series of cases reported from America and Egypt, one gathers the impression that the condition must be very much more common in these lands. However, two cases of nasopharyngeal fibroma have been seen and treated in this department within a year with one other case in another hospital and it, therefore, seems appropriate to review briefly the more recent literature on the subject, and describe the treatment of these cases.

Ætiology

There seems to be an overwhelming opinion that the condition is a true neoplasm, although Willis (1953) will not accept this. He maintains that true fibroblastic tumours occur only rarely in the nasal or nasopharyngeal mucosa, and that most so-called tumours in these regions are really inflammatory or allergic overgrowths. Their tendencies to recur, to spread over a wide field of diseased mucosa, and to lead to infective complications in adjacent cavities, bones and tissues have, he says, "given these a false clinical reputation of neoplastic qualities". However, Willis does not completely deny the existence of true fibroblastic tumours in this region, and this rarity of both pathological and clinical condition seems to suggest that the fibroma may still be truly neoplastic.

Age

Negus (1956) while giving their cause as obscure, suggests that they may arise from developmental irregularities at puberty. Cases have been reported, however, which have attended with the established condition at a younger age. Both the cases seen here, were prepubertal as was also Case III. Handousa (1954) reported a series of 70 cases, the age on admission varying from 8 to 50 years. He states that the disease is predominant at, but not limited to, puberty. Martin (1954) reported a series of five cases with an average age of 17 years. Martin, Ehrlich and Abels (1948) gave an average age of 16 in a series of 29 cases Figi and Davis (1950) presented 51 cases with an age average of 15 1 years, and Harkönen and Malmio (1948) had an average of 16 5 years in their series of 31 cases Martens and MacPherson (1956) reporting a series of 10 cases from the files of the Armed Forces Institute of Pathology, found an age period ranging from 13 years to 24 years with one patient aged 55 years

Sex

Some, even recent, textbooks on Diseases of the Ear, Nose and Throat state that the condition is exclusive to males This is also the belief of Hayes Martin (1948) who links this to the common onset at puberty to postulate a hormonal theory of origin He believes that there is a deficiency of androgens, or a surplus of œstrogens producing a hyperæmia of the mucosa from which the tumour eventually arises To support this theory, he cites two other findings One is sexual underdevelopment in this condition—this has been refuted by other workers—the other is the tendency for these tumours to regress after the age of 25 years This latter observation has also been contested (Handousa, 1954) and also by others Most authorities, however, while agreeing that the condition predominates in males do not categorically deny that it can exist in the female Figi (1940) reported the condition in five females in his series of 63 cases Handousa (1954) reported it in 11 females in his series of 70 cases This author also categorically denies having seen, in his large series, any case which showed regression after the age of 25 years, or indeed, any case which showed sexual underdevelopment These observations, supported by other writers, seem to undermine the validity of the hormone theory of origin in fact J S Martin (1954) goes so far as to state that "sex hormone unbalance may have some effect on the vascularity of these tumours, but it cannot be responsible for their initiation, nor for determining their location in the nasopharynx '

Site

It is generally accepted that the commonest site of origin is from the periosteum lining the vault of the nasopharynx, usually from the lateral wall above the opening of the Eustachian tube, but also from the roof or the posterior wall at the level of the upper cervical vertebrae A theory has been propounded that the site of origin is from the perichondrium of the embryonic cartilage between the basisphenoid and basiocciput This cartilage ossifies at about 25 years of age This has been offered as a reason for the spontaneous regression stated to occur after the age of 25 years However, there is no conclusive proof to support this

Pathology

Macroscopically, the tumour is usually smooth or lobulated, firm to palpation but not stony hard. The colour varies from pale pink to deep red. The surface may be ulcerated or sloughing, particularly after hæmorrhage or surgical manipulation, while occasionally the tumour mass may be distinctly friable. In size there is great variation, from purely nasopharyngeal to massive extensions downwards below the soft palate, or forwards into the nose, ethmoids, antrum, orbit and cheek, or upwards into the cranial cavity. The route of extension into the cheek is interesting. Handousa (1954) asserts that, in all tumours showing this extension, the tumour "wound round" the outer border of the maxilla and not through it. The first of our cases had most likely extended into the cheek by this method and also through the antrum by erosion of the posterior and lateral antral wall.

Rate of growth is likewise as variable as the size of the tumour. Scott-Brown (1952) states that it may be so rapid as to suggest sarcoma. Certainly, one of the cases reported here showed such comparatively rapid growth that a diagnosis of sarcoma was at one time much more favoured than one of fibroma, despite negative pathological reports of malignancy. Pressure atrophy of bone and neighbouring structures is also a feature of these tumours. Malignant change with invasion has never definitely been reported, nor secondary metastases, though Batsakis *et al.* (1955) reported a case of a male, aged 54 years, who had undergone two surgical operations for removal of a nasopharyngeal fibroma, in 1949 and 1952. This was followed by irradiation for recurrence in 1954, a total of 9,000 r being given, with large doses of androgens. A third removal was done in 1954, and histological examination of the operation specimen revealed areas of fibrosarcoma. It is possible that malignant change had supervened de novo, but also that it arose following radiotherapy, so-called "radiation cancer".

Microscopically, the tumour is a vascular fibroma, in parts so vascular as to be termed an angiofibroma. There is no true capsule, the tumour having an epithelial cover, either squamous or, in parts, pseudo-stratified columnar epithelium. The surface may, however, show atrophy or ulceration, and sub-epithelial infiltration of plasma cells, lymphocytes and polymorphs is not uncommon. The blood vessels are said to be of two types, firstly, normal vascular channels with muscular coats, and secondly, primitive blood vessels without elastic tissue in the walls. Brunner (1942) states that "angiomatous tumour vessels communicate with the veins of surrounding tissue only". From this, he argues that carotid ligation as a preliminary to surgical removal is not indicated, nor effective. Hyaline degeneration of the vessel walls may cause thrombosis, with necrosis of areas of the tumour and subsequent shrinking. This may explain the diminution in size noted on occasion and perhaps ascribed to regression of the tumour Myxomatous degeneration has also been noted as a feature of these tumours

Clinical Features

Current textbooks are unanimous in two outstanding clinical features of this condition, namely, nasal obstruction and epistaxis The former is progressive and very often becomes unilaterally or indeed bilaterally complete Epistaxis is usually intermittent, and may be slight or most alarmingly profuse and spontaneous It is almost always heavy following surgical manipulation of the tumour

Other features are due to extension of the tumour, an early symptom being deafness of a conductive type, following interference with the patency of the Eustachian tube Oddly enough, actual aural suppuration is rare Nasal and nasal sinus suppuration, facial deformities, interference with ocular movement, dysphagia and anæmia of microcytic type are other features Cranial nerve palsies from tumour extension are not as common as might be supposed, and one other rather surprising finding is often the absence of pain

Diagnosis

This rests upon the gradual history, appearance and consistency of the tumour, and absence of evidence of secondary metastases in regional lymph glands Additional aids to diagnosis are radiological examination and biopsy The former will show the tumour as a soft tissue mass occupying the nasopharynx, with evidence of extension into the nose when present and perhaps also destruction of bone, in particular the maxilla The latter procedure is looked upon with disfavour, perhaps on account of the very severe bleeding which may attend the removal of a biopsy specimen, but it is not impossible to do if adequate precautions are taken Diathermy removal of a specimen has been advised to counteract the bleeding tendency However, the biggest failing in a biopsy specimen is that it may be difficult to get one which is truly representative

Differential Diagnosis

This rests between fibroma and other space occupying lesions of the nasopharynx

(1) Adenoidal hypertrophy—in this condition the obstruction to breathing is unlikely to be so profound as in fibroma, while epistaxis is certainly much less frequent and extensive The appearance of the vertically lobulated mass on posterior rhinoscopy is almost diagnostic in the child and adolescent, but may cause a little more difficulty in the adult in that it is less expected

(2) Choanal polypi—are generally soft, whitish, translucent and pedunculated, but if subject to infection and fibrosis, may assume more the appearance of a fibroma. Polypi may also be present in the nose, while radiological evidence of maxillary sinus infection is almost always present.

(3) Carcinoma—this condition occurs at a much later decade in life than fibroma. Epistaxis may often be a feature without being of massive proportion and pain is usually present, but very often the first indication of the disease is the appearance of secondary metastases in the cervical lymph nodes. Direct invasion particularly of the base of the skull with cranial nerve involvement, in particular the third, fifth and sixth and latterly the nerves of the jugular foramen, are also features. Clinically, the growth is often small and granular or ulcerated, and stony hard. Indeed it may be very difficult to see, especially if it is sub-mucosal. A biopsy will indicate its true nature.

(4) Sarcoma—this tumour, more common in the younger age groups, usually shows rapid growth, with early metastases and cachexia. Ulceration is not an early feature.

(5) Other tumours—include simple fibroma, chordoma, plasmocytoma, chondroma, scleroma and occasionally ectopic salivary gland tumours. Dermoid cysts, teratoma, Thornwaldt's bursitis and retropharyngeal abscess cause rare swellings in the nasopharynx. Diagnosis must be by clinical appearance, consistency, and site, with radiological appearances and biopsy.

Treatment

The fear of hæmorrhage has in the past tended to make the surgeon hesitate to advise surgical removal. On the other hand, these tumours may become so extensive that the oft-described regression which is said to occur around the age of 25 years, can hardly be awaited. Several non-surgical or minor surgical techniques have, therefore, been tried. Briefly, these are:

(1) *Injection of sclerosing solutions.* Forty per cent. saline injected in quantities varying from 3 to 10 cc. into various parts of the tumour at intervals of 2-3 weeks have been recommended (Watkyn-Thomas).

(2) *Diathermy removal or acupuncture,* again usually at intervals and with several applications.

(3) *Sex-hormone therapy*—Martin (1948) considers there is a deficiency of androgens, and advocates androgen replacement therapy to correct this.

(4) *Radiotherapy.* This method has been widely practised in America especially at the Mayo Clinic. The radiotherapy may be

given as deep X-ray, or by implantation of radon seeds or radium needles, and sometimes combined with diathermy coagulation Treatment is necessarily protracted and aims at reducing the size and vascularity of the tumour Good results are claimed, although Handousa (1954) states that he has "never seen a nasopharyngeal fibroma that has completely disappeared after any type of irradiation" Moreover, in young children, the possible detrimental effect of radiation upon centres of ossification should be borne in mind

Various surgical approaches have been devised, and it suffices to name the more popular routes only These are

(1) Moure's Lateral Rhinotomy

(2) Denker's sublabial approach

(3) Transpalatal approach (Ruddy, 1945, Wilson, 1951)

The aim of all these procedures is to expose and mobilize the tumour so that its attachments may be severed subperiosteally, i e between the tumour base and the underlying bone In this way, many of the vascular channels remain undamaged Former methods of avulsion and morcellement were usually attended by massive hæmorrhage, and may have contributed to the dread attending surgical intervention Post-operative shock is usually severe and necessitates the replacement of blood by transfusion, in fact, pre-operative transfusion is often beneficial and counteracts the tendency to microcytic anæmia which most of these patients show

In two of the cases about to be described, the approach was by lateral rhinotomy in the first case, and by the transpalatal route in the second The first case was also undertaken with the aid of one of the newer techniques in anæsthesia, the induction of a state of hypothermia From the surgical viewpoint, there appeared to be considerably diminished bleeding during operation, and the immediate post-operative shock was very much less severe than anticipated So far as I am aware, this technique has not been reported in the surgery of nasopharyngeal fibroma to date

CASE I

B M , male, aged 11 years First attended the E N T out-patient clinic on 24 2 56, with a complaint of intermittent nasal obstruction, not complete, with slight mucopurulent discharge, for approximately 6 months There was no pain, nor any bleeding from the nose He had had his tonsils and adenoids removed approximately 3½ years previously Examination revealed hypertrophic inferior turbinals on both sides of the nose, with what was thought to be some adenoid regrowth in the nasopharynx A radiograph of the nasal sinuses showed some clouding of the left antrum He was treated conservatively for a month with Argyrol and

Ephedrine nasal drops. When seen again on 6.4.56, there had been little improvement, and as a good view of the nasopharynx was difficult to obtain, admission for examination under anæsthesia was recommended. 18.5.56. Examination with Xylocaine local anæsthesia—both nostrils were occluded; on the left a firm pink swelling filled the middle meatus and was pushing the middle turbinal upwards; on the right, a similar swelling was seen far back in the choana. Posterior rhinoscopy showed the nasopharynx to be filled with a firm mass of tissue, pink in colour and quite smooth, and completely occluding the choanae. In the mouth, there was an outward bulging of the antro-lateral wall of the left maxillary antrum, with the upper VIIth molar lying horizontally and causing a contact ulcer on the inner surface of the left cheek. There were no palpable glands, eye movements were good with no diplopia. Radiological examination showed

Fig 1

Case I Radiograph showing an opaque left antrum, with some displacement of the medial antral wall to the right The lateral antral wall is thin and eroded A soft tissue mass has extended laterally beyond the left maxilla and also obscures both nares

(Figs 1 and 2) an 'opaque left antrum The lateral antral wall is very thin with an erosion P A view shows some displacement of the medial wall of the left antrum to the right There is a soft tissue mass occupying most of the nasopharynx A specimen was taken from the nasopharynx for pathological examination on 23 5 56 There was quite marked bleeding following the removal of the tissue which was adequately controlled with a nasopharyngeal pack The pathologist reported the specimen as showing ' regular epithelium with chronic inflammatory changes in the sub epithelial tissues No evidence of a malignancy In view of this a further biopsy was taken a week later, and this was reported as adenoid tissue with chronic inflammatory change No evidence of malignancy Repeat X ray examination while confirming the previous findings also reported decalcification of the left upper alveolus The displaced upper left molar

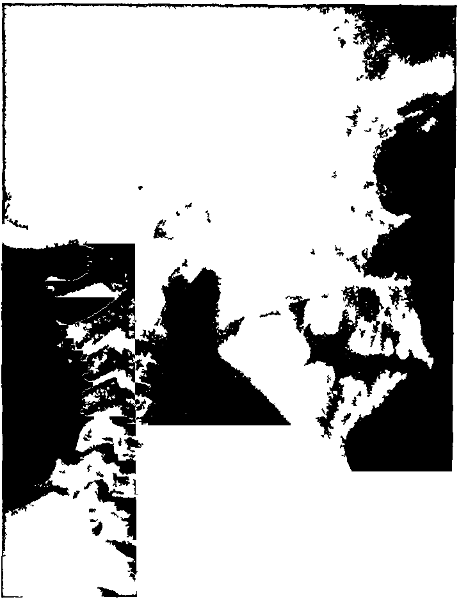
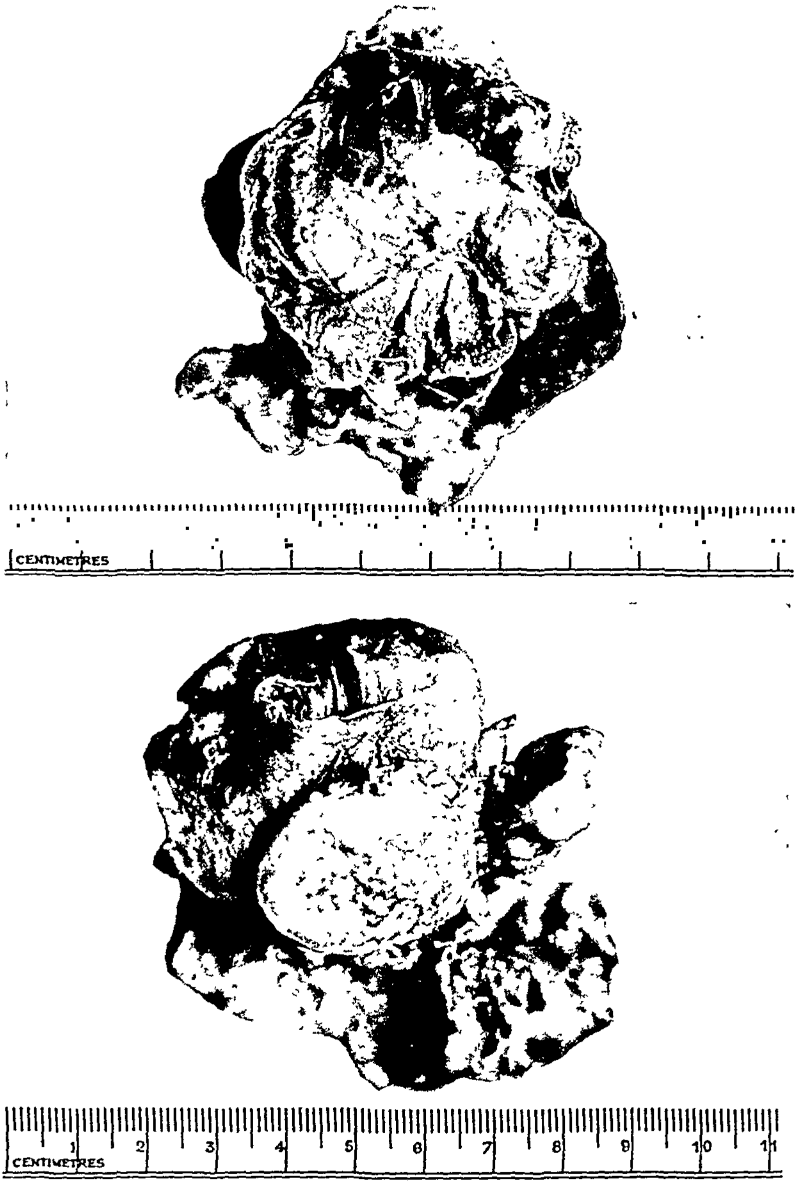


FIG 2

Case I Lateral radiograph showing the nasal cavity and nasopharynx occupied by a soft tissue mass



FIGS. 3 and 4.
Case I. Gross appearances of nasopharyngeal fibroma, showing the tumour size and extensions.

He now began to have frequent and severe attacks of epistaxis, a symptom noticeably absent on the first occasion.

A repeat biopsy was again taken, and histological confirmation of nasopharyngeal fibroma again obtained. The obvious extent of the tumour recurrence with the possibility of wide excision for removal made it desirable on this second occasion to seek the advice of the plastic surgeon. The boy unfortunately developed an acute appendicitis on 8.3.57, necessitating appendicectomy and a delay of two weeks.

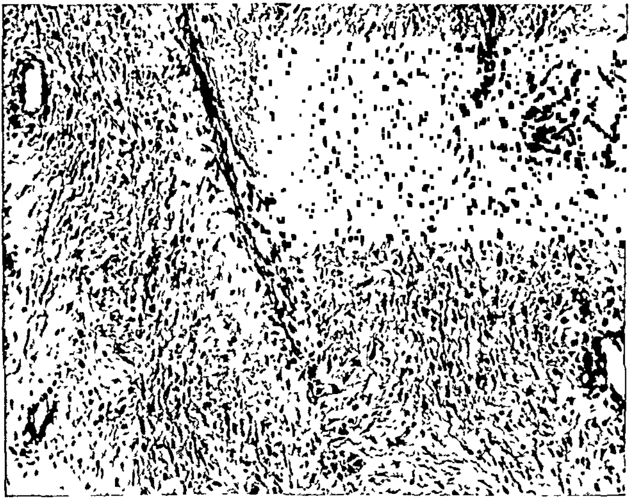


FIG 5

Case I H and E ×240 Showing masses of young fibrocytes with fibrils Numerous capillary blood vessels with thin walls

The considerations in treatment in this case now were:

(1) This was a histologically simple tumour and, therefore, the ideal treatment should be adequate exposure and local removal.

(2) The tumour had again assumed massive proportions. In such cases, the tumour may have secondary attachments and, therefore, limited exposure through the nose and mouth seems to have a high recurrence rate. Such limited exposure, except in early cases, must be difficult and unsatisfactory, with danger from hæmorrhage.

After consultation and examination by the plastic surgeon, it was decided that, as the recurrence was obviously involving the orbit and ethmoids, a complete removal of maxilla and ethmoid with exenteration of the left orbit offered the best chance of completely removing this tumour.

Operation was carried out on 24.4.57, using a hypotensive technique with anæsthesia.

The approach was by a modified Ferguson incision—the original lateral rhinotomy incision extended through the upper lip, and laterally along the lower border of the orbit to the outer canthus. The modifications employed were:

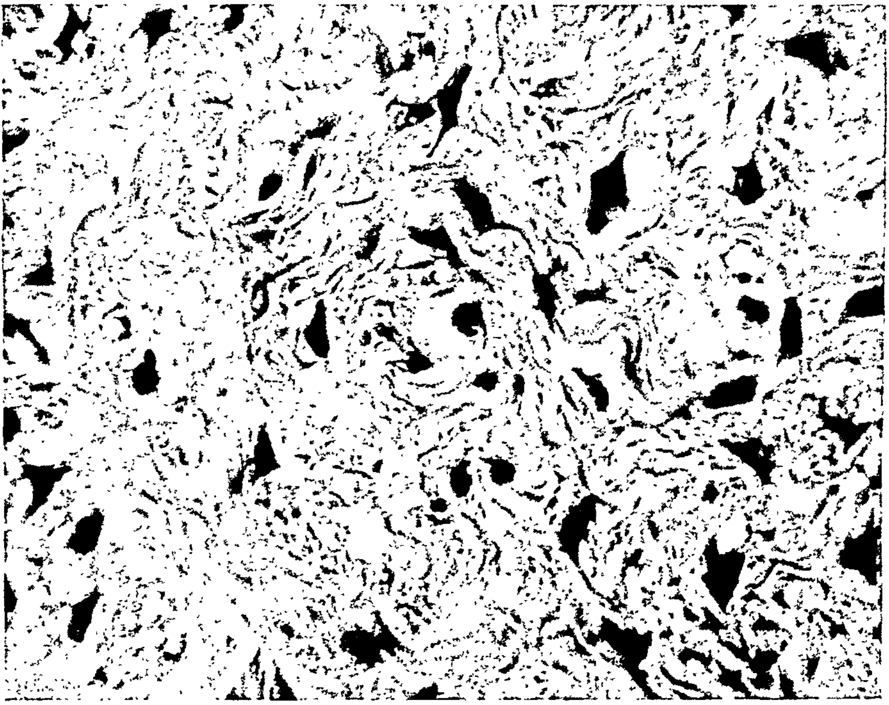


FIG. 6.

Case I. H. and E. × 880. Showing fibrocytes with the dense interweaving of connective tissue fibrils.

(1) The incision in the upper lip was angled. This prevents subsequent notching after suture.

(2) The incision in the lower eyelid is kept at 3 millimetres from the lid margin. This prevents troublesome œdema of the lower lid, which often follows the older incision along the intraorbital margin.

The left cheek was widely reflected laterally, readily exposing the tumour, which was seen to be filling the left side of the nose and the antrum, to have eroded the posterior wall of the antrum and the orbital floor and to have extended behind the globe. The main mass, with maxilla and orbital contents, was removed complete. It was seen on removal to have a broad attachment to the posterior nasopharyngeal wall and vault and to have laid the sphenoid sinus open. A split skin graft was then used to line the raw surface of the cheek. This prevents contracture, allows inspection of the cavity, and accommodation of an internal prosthesis for support.

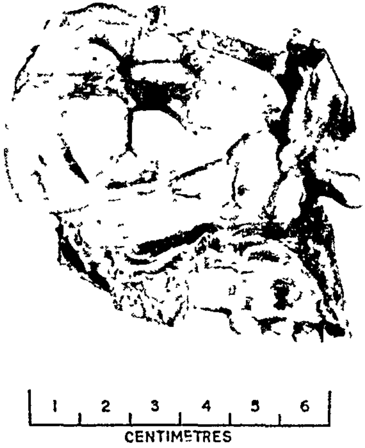


FIG 7

Case I Gross appearance of tumour and maxilla removed en bloc The tumour has completely eroded the posterior maxillary wall and occupies the antrum entirely.

Recovery from the operation was uneventful. An internal prosthesis was fitted and changed at regular intervals, when adjustments were made and the cavity inspected. At a later date an external eye prosthesis was made. Apart from some nasal bleeding, due to the atrophic condition of the nasal mucosa on the left side, the patient has remained well and free from tumour to date.

Examination of the operative specimen (Fig. 7) showed it to be a simple nasopharyngeal fibroma, of identical pathology to the original tumour.

Case II was again a boy aged 10 years. He was first seen on 3.6.57, when the complaint was of recurrent epistaxis for approximately three months, mostly from the right nostril. This had been particularly severe for two days prior to examination. The right nostril was partly obstructed but not completely so. There was no history of trauma to the nose, nor of a familial bleeding tendency. Pain was absent, and deafness was not a feature.

Examination of the nose revealed a pinkish mucosal projection rather far back in the right nostril, in the inferior meatus, with what looked like a fresh bleeding point on the projection. This latter was very similar in appearance to a hypertrophied posterior end of inferior turbinal. Application of the electro-cautery to the suspicious area rather aggravated than controlled the bleeding, and the nose was packed firmly with ribbon gauze soaked in a liquid paraffin/penicillin/sulphamezathine preparation. This packing controlled the bleeding and was left *in situ* for 48 hours. Hæmatological examination at this time gave the following information:

Hæmoglobin 80 per cent.
Red blood cells 4·2 million/c.mm.
White blood cells 8,000/c.mm.
Packed cell volume 34 per cent.

Blood film showed no significant abnormality.

On 5.6.57 the packing was removed. Bleeding recommenced and again was not adequately controlled by cautery. A biopsy from the mucosal projection in the right nostril was taken and the nose again packed, with control of the bleeding. Histological examination of the biopsy specimen showed blood and pus cells only, with some mucosal surface epithelium. Radiological examination of the chest was negative, but the nasal sinus view showed opacity of both maxillary antra.

10.6.57. Bilateral antral lavage under general anæsthesia produced pus from both antra. Palpation of the nasopharynx revealed the presence of a smooth, firm swelling which appeared to be attached to the upper posterior wall of the nasopharynx. Examination of the right nostril again showed the firm mass in the posterior half. Biopsies were taken from both the nasopharyngeal mass and again from the right nostril, the bleeding being quite easily controlled by pressure. Polythene tubing was introduced into both antra. Culture of pus from the antra grew Gram positive diplococci, *Staphylococcus aureus* and β-hæmolytic streptococci, sensitive to Chloromycetin and Streptomycin.

Histological examination of both biopsy specimens showed them to consist of "moderately cellular fibrous connective tissue, covered by stratified columnar epithelium. Blood vessels are very numerous, many having very thin walls and being of considerable size. The appearance is consistent with a juvenile nasopharyngeal fibroma" (Fig. 8).

Bearing in mind the experience gained with the previous case, surgical treatment was decided upon without delay. In this case the tumour was very considerably smaller, and without any extension other than partly into the right nostril. It was felt that a transpalatal approach similar to that suggested by Ruddy or Wilson would give adequate exposure.

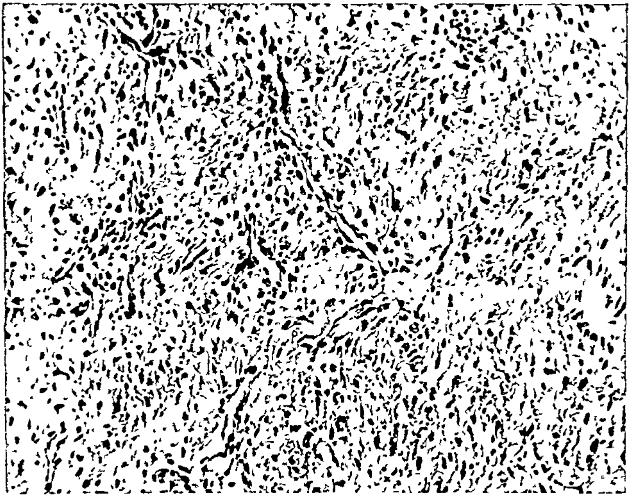


FIG 8

Case II H and E × 240 Showing many fibrocytes and connective tissue fibrils Blood vessels are very numerous and thin-walled

14.6.57. Removal of the tumour through a Wilson (transpalatal) approach. Anæsthesia was obtained by injection of thiopentone 0·4 gm. and Scoline 40 mgm., and maintained with nitrous oxide and oxygen through a per-oral endotracheal tube. An intravenous saline drip was started, containing "Arfonad" 375 mgm.—as a 0·1 per cent. solution—

and by this means a substantial fall in blood pressure was obtained. The tumour was seen to have a fairly broad and strong attachment to the roof of the nasopharyngeal vault and to a lesser extent, the lateral wall. The nasal prolongation was lying unattached in the right nostril, and was easily manipulated backwards into the nasopharynx. The whole mass was then removed by dissection close to the bony wall of the vault. The tumour appeared almost encapsulated. The cavity was then packed with B.I.P.P. gauze. Bleeding was minimal throughout.

FIG 9

Case III. Radiograph showing a soft tissue mass occupying the left nasal cavity and expanding the lateral nasal wall laterally and the nasal septum to the right

The packing was removed the next day without further bleeding. On 28.6.57 both antra were again irrigated and found to be clear. There was a small perforation in the incision line at the junction of the hard and soft palates, but one month later this had healed The nasopharynx was quite free from tumour, and had remained so at the last visit on 3 12 57

The third case was first seen at the Royal Hospital for Sick Children in Aberdeen on 13 4 57, having been flown there from Orkney as a case of fractured nose with increasing nasal obstruction, of seven weeks duration Clinical examination revealed a deviation of the cartilaginous nasal septum to the right side, with granulation tissue occluding the left nostril The soft palate was slightly depressed on the left side Posterior rhinoscopy revealed a round pale pink mass filling the nasopharynx, and having a large vein upon its surface The right eardrum was healthy, the left was indrawn, with a negative Rinne result upon testing the left ear with the tuning fork Externally, there appeared to be a soft tissue swelling presenting as an "hour-glass" deformity above and below the left zygomatic arch, and causing fullness over the left parotid

Radiological examination revealed the following information

"Skull, including view of the left orbit there is a large soft tissue mass involving the left nasal cavity posteriorly which is expanding the lateral wall of the nose in the region of the pterygoid laminae and posterior maxilla The septum is deviated to the right Basal view shows a probable erosion of the pterygoid laminae with extension of the soft tissue mass into the temporal fossa, probably through the pterygo-maxillary fissure The mass would appear to be mainly in the nasopharynx and posterior choana Appearances are those of a tumour, causing displacement by expansion rather than by erosion, although there is some destruction at the lower end of the septum" (Figs 9 and 10)

To confirm the clinical findings, examination under general anæsthesia was carried out on 16 4 57 Palpation demonstrated a smooth, hard mass in the nasopharynx, which appeared to arise from the postero superior wall and to the left side A biopsy was taken from the presenting nasal portion, and histological examination of this showed "pyogenic granulation tissue"

Although at this stage a definite diagnosis of nasopharyngeal fibroma had not been made clinically the evidence was strong in favour of this The choice of treatment lay between surgery and radiotherapy, and after consultation, it was decided to give precedence to the latter method This was begun on 18 4 57 The first course lasted until 3 5 57 Two opposed fields 10 cm by 10 cm were used, with a separation of 14 cm Treatment was given at 250 kv with a triple Thoreaus filter to a central dose of 1,500 r, the maximum dose being 1,680 r He was given 10 treatments

8 5 57 Biopsy with diathermy of a large part of the nasal mass Histologically this was reported as "cellular and highly vascular granulation tissue showing much superficial ulceration and infection In some areas the fibroblasts are large and aberrant in form, and mitotic activity is slightly excessive This lesion cannot be diagnosed as true neoplasia, but it seems

to conform to the pattern of a juvenile nasopharyngeal fibroma which tends, for a time at least, to be of locally infiltrative behaviour."

The response to radiotherapy was encouraging, considerable reduction in the size of the tumour mass being evident. In view of this, a second course, identical to the one described, was given, from 26.6.57 until 11.7.57.

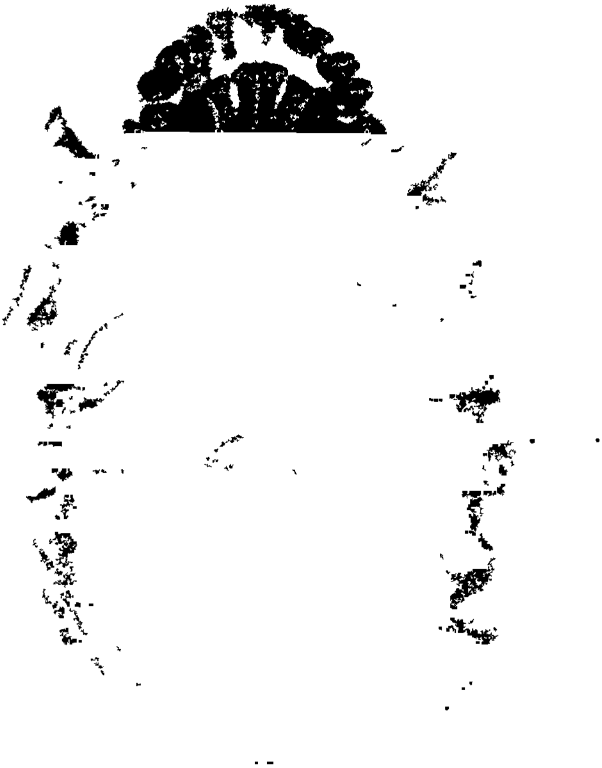


FIG. 10.

Case III Basal view showing a probable erosion of the pterygoid laminae on the left, with extension of a soft tissue mass into the temporal fossa. L.

Improvement continued to be maintained, with steady regression of the tumour, and when seen on 1.12.57, the post-nasal and intra-nasal mass was lessened by 50 per cent. When seen again on 6.3.58, the post-nasal tumour had shrunk down to a small pedunculated hard mass about one-half inch in diameter, arising from the nasopharyngeal vault. Inspection of the left nostril showed the tumour to have receded to the posterior nares. X-ray examination (Fig. 11) shows the extent of improvement.

Summary

1. Three cases of nasopharyngeal fibroma are described.

2. The treatment of these cases is described, and indicates the merits of both surgical and non-surgical forms of treatment.

3. The employment of the newer anæsthetic techniques of hypotension and hypothermia has been shown to materially lessen the danger of hæmorrhage and shock in surgery of nasopharyngeal fibroma.

4. The more recent literature on the subject has been reviewed.

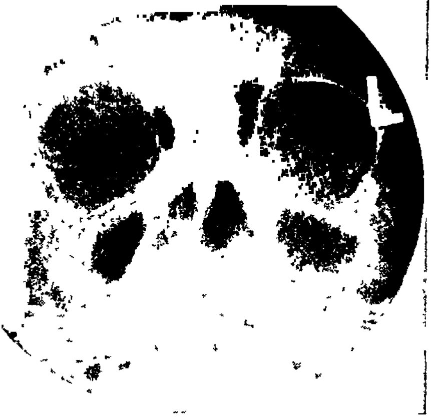


FIG 11

Case III. Radiograph showing reduction in size of soft tissue shadow following radiotherapy (cf Fig 9)

Acknowledgments

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TERATOID TUMOURS OF THE NASOPHARYNX

By P B FOXWELL and B H KELHAM (London)

Introduction

TERATOID tumours of the nasopharynx form a rare but well-recognized group of tumours A study of the records at King's College Hospital as far back as 1928 has failed to reveal a single case, and the recent occurrence of two cases within seven months of each other stimulated our interest in the subject

Review of Literature

Teratoid tumours of the mouth, pharynx and nasopharynx have naturally been of great interest to the teratologists, and a study of the literature shows that the main contributions on the subject have been by them Thus there were important papers by Ahlfeld (1875), Arnold (1888), Marchand (1897) and Schwalbe (1907) Ehrich (1945) surveys the literature relating to teratoid tumours of the pharynx

Probably because of the rarity of cases in clinical practice, the subject has aroused little interest among oto laryngologists, who have mainly been content to record cases occurring in their practice as interesting curiosities The first, and indeed the only notable contribution in the English literature was by Brown Kelly (1918), who confined his attention to dermoid cysts of the nasopharynx—the so called hairy polyps Oppikofer (1932) reviewed 62 hairy polyps, di Vestea (1937) reviewed the French and Italian literature on the subject

Classification

Ewing (1940) recognizes four main types (*a*) Dermoids They appear at birth or later, and are attached to the hard or soft palate, or wall or vault of the pharynx, in or near the midline, and fill the nares or buccal cavity Some show an intra cranial portion connected through a perforation in the skull with the pharyngeal tumours

The polyps are covered by skin, often hairy, and hence are often called hairy polyps They contain dermal glands The main mass is composed largely of fat tissue, with fragments of striated muscle, cartilage, or bone They are, therefore, bidermal (*b*) Teratoid tumours and (*c*) Teratomas. These differ from the dermoids chiefly in the greater complexity of structure, larger size and earlier development. They are frequently associated with extensive deformities of the skull, as anencephalis, hemicrania, and palatal fissures. These tumours are tridermal, and in the teratomas the three germ layers produce definite organs.

The tridermal elements consist of fat, muscle, cartilage, bone, acinous glands, teeth, cysts lined by various forms of epithelium, and nervous tissue.

(*d*) Epignathi. These tumours present well-formed organs and limbs of a parasitic fœtus.

Comments on the Nature of the Tumours

(1) *Old Views.* The basis of former views on the ætiology of teratomas is embodied in the definition given by Bland-Sutton (1922): A teratoma is an irregular conglomerate mass containing tissues and fragments of viscera belonging to a suppressed fœtus attached to an otherwise normal individual.

Arnold favoured a dual origin for the tumours. The dermoid cysts and simpler tridermal growths were thought to be due to local disturbances of development involving the buccal cavity, while the true teratomas (presenting rudimentary organs) were true fœtal implantations, and due to the aberrant growth of a totipotent blastomere, polar body, sex cell or tissue bud.

It was soon realized that this dual theory did not fit the observed facts, since the sharp separation between the two groups is impossible. Other pathologists attempted to explain the origin of all the tumours by one mechanism. Thus Ahlfeld thought they were due to twin formation; Schwalbe that they were due to dislocation of germ material.

These ideas were generally felt to be unsatisfactory, and have been well criticized by Askanazy (1907) and Nicholson (1950). The whole concept that these tumours represent an attempt, with varying degrees of success, to form a new individual has been abandoned.

(2) *New Views.* Recently the view has been gaining ground that a distinction must be drawn between congenital malformations and true tumours. It is believed that beneath their superficial similarity there are two distinct kinds of teratoids—the first, a malformation and the second, a neoplasm (Payling Wright, 1954). According to this view, a teratoma is defined as a "true tumour or neoplasm composed of multiple tissues of kinds foreign to the part in which it arises" (Willis, 1953); as such it differs from the teratoid masses, which are congenital malformations.

The teratoid masses arise close to the midline axis of the body; examples are the present group of tumours of the nasopharynx; sacrococcygeal teratoids; intrathoracic teratoids; retroperitoneal teratoids. The commonest examples of teratomas are the teratomas of the gonads.

Payling Wright differentiates these abnormalities as follows. (1) There are the teratoid masses which arise from some developmental error in early embryogenesis, are usually apparent at birth or shortly after, and grow more or less concurrently with the bearer. Possibly they are caused by a cell or group of cells, which still possesses wide powers of differentiation, becoming separated physiologically from the broad forces that govern the orderly development of the embryo, and in consequence undergo further differentiation solely under the influence of its own intrinsic determining factors. Such growths are not true tumours and should be classed among the malformations or complex hamartomas.

Both of the cases which we shall describe later would come into this category.

(2) There are true neoplasms which take their origin in these malformation teratoids; the malignant change may occur at any period in the life of a teratoid from infancy to old age.

(3) There are the teratomas which arise in the gonads, usually in middle life, and in the testis assume a highly malignant form. It seems established that teratomas can arise as a result of an exogenous irritant in testes that were originally free from any congenital abnormality.

Frequency of Occurrence

Hairy polyps are much the most frequent. The first recorded case was in 1784. By 1905 20 cases had been recorded; by 1911 40; and Brown Kelly found 50 by 1918. Ehrich in 1945 brought the number up to 75. Kesson found 12 additional cases by 1954, including his own. References to two further cases have been found, making the total 89.

From the viewpoint of the otolaryngologist, the more highly developed teratoid tumours are much rarer, since in general the more highly developed the tumour, the greater the chance of a still-born fœtus. Thus Howarth, describing a teratoid tumour in 1938, stated that he had only been able to find six similar cases.

Age and Sex

More than half of the dermoid cysts occur in infants under 1 year of age, and the majority of these very shortly after birth. The rest may present at any time up to adult life, and Ehrich mentions a case aged 66 years. It is rare, however, for a case to present after 21 years. Of 22 higher developed parasites mentioned by Arnold (quoted by Ehrich), 14 occurred in fœtuses, 5 in newborn infants, and 3 in infants that died 1-5 days after delivery.

As regards sex incidence, the tumours occur much more commonly in females, in the proportion of 6-7:1. Of the two cases described below, one was male, the other female.

Clinical Aspects

The clinical picture will naturally depend on the size and situation of the tumour. Thus, in a few cases the infant has been born with the tumour protruding from the mouth. Again, the tumour may be of such a size or so situated as to cause partial asphyxiation and demand immediate removal. The majority of hairy polyps are pedunculated, and not infrequently the pedicle is long enough to allow the tumour considerable mobility. In such cases symptoms will be intermittent, with attacks of coughing and cyanosis if the tumour comes to lie over the larynx, and dysphagia. Sometimes the mother will mention seeing the tumour after a bout of coughing has expelled it into the mouth. In Bulson's case (1926) the child had occasional paroxysms of suffocation and was relieved by being held upside down and patted on the back.

Sessile tumours, or tumours with a very short pedicle, which completely obstruct the nasopharynx will cause severe symptoms, not only because of the obstruction to the normal nasal airway but also because the mass of the tumour may impede mouth-breathing as well. Asphyxiation is, therefore, a much greater danger than in bilateral choanal atresia, and feeding difficulties are also greater. The usual signs of respiratory distress, including cyanosis, intercostal recession and supraclavicular recession, are to be expected. There may be diminished air entry into the lungs. Where the nasopharyngeal obstruction is only partial, symptoms will be correspondingly milder, and it may be some weeks or months before the child is brought up for examination. In such cases snoring and snuffling are commonly complained of; an excessive amount of mucus may be present in the nose; feeding may be noisy and prolonged; there may be failure to gain weight satisfactorily.

Small tumours may escape diagnosis altogether in the absence of symptoms, and some are discovered accidentally during routine examinations for other conditions.

Diagnosis

A study of the literature reveals that there has rarely been much difficulty experienced in the diagnosis of the larger tumours. With the pedunculated tumours, the mass or its pedicle is usually seen on oral examination. Sometimes, when nothing is immediately obvious, the tumour will become visible when the child coughs or gags during examination. Sessile tumours can usually be seen projecting below the soft palate, and the soft palate itself may be bulged downwards and forwards.

The only cases likely to give rise to difficulty are those where the growth lies entirely within the confines of the nasopharynx, and particularly where the nasopharyngeal obstruction is not complete. In such cases we would like to emphasize the value of a lateral X-ray of the nasopharynx. It may be particularly valuable in differentiating the condition from choanal atresia. The X-ray taken in our second case shows the bony and soft tissue mass and illustrates how both nasal and mouth breathing are obstructed (Fig. 1).

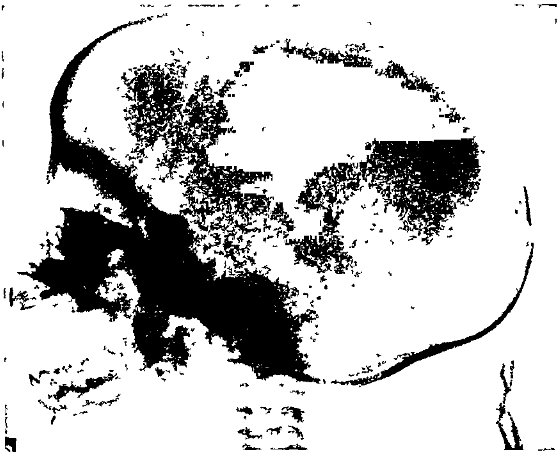

FIG 1

Lateral X-ray of Nasopharynx (Case II) Showing depression of soft palate by the bony and soft tissue mass (The shadows in the anterior nares are caused by lipiodol)

As mentioned above, other developmental anomalies are rare with hairy polyps, but the chances are much greater with the more highly developed tumours.

In cases seen later in life, these tumours are often mistaken for fibromas or similar growths until examined histologically after removal.

Prognosis

As has been made clear above, this will depend on the histological development of the growth and on the presence of associated deformities. It is a rather surprising fact that in all the cases of hairy polyps, not a single death has been recorded so far as can be ascertained. Neither has there been any report of a recurrence.

Treatment

The immediate treatment will depend on the severity of the symptoms, and on whether the tumour is sessile or pedunculated. Operation is difficult in day-old infants, and in the case of sessile post-nasal growths should be delayed if the child seems likely to make satisfactory progress. Pedunculated masses are easier to deal with and should be removed.

With regard to anæsthesia, many tumours diagnosed in infancy have been removed without any form of anæsthesia. However, unless the urgency of symptoms is such that immediate removal is necessary, our preference is for general anæsthesia, because firstly, this avoids operating upon a struggling patient; secondly, the mouth can be adequately opened; and thirdly, hæmorrhage can be controlled under direct vision. Naturally general anæsthesia in infants, especially where there is already some respiratory obstruction, should only be undertaken by a skilled anæsthetist. The difficulty in maintaining a good airway, especially during induction, may be considerable. This was so in our first case. The exact form of anæsthesia will, of course, vary with the anæsthetist, but the methods used in our two cases are described below.

It is essential to have a tracheostomy set at hand in these cases, though it is unlikely to be needed. Tracheostomy is to be avoided if possible, because of the danger of interference with the later growth of the larynx; difficulty of decannulation in infants; and the risk of subsequent stenosis of the larynx.

The method of removal of the tumour will depend on the type of growth. If it is pedunculated, then removal with a snare is the best method, and has been used in the majority of cases of hairy polyps. If the tumour is sessile, as it was in our two cases, it may be seized with grasping forceps passed behind the soft palate, and avulsed. This is not an easy manœuvre in these tiny patients, but more elaborate methods are not likely to improve results. In our cases, the amount of bleeding after this procedure was remarkably slight; although brisk at first, it stopped very quickly and was certainly no greater than after adenoidectomy.

Case Reports

CASE I. This was a full-term, first-born male child born on 26.3.57, after a normal pregnancy and labour. The birth-weight was 6 lb. 3 oz. Respiratory obstruction and feeding difficulties dating from delivery had been noted, followed by mucoid discharge from the right nostril.

The baby came under the care of Mr. Lewis when it was 10 days old, 5.4.57. It was a weakly, ill baby whose pink colour was barely maintained in the oxygen tent in which it arrived. Since suckling precipitated acute respiratory obstruction, 4-hourly tube feeding since birth had been necessary, and the weight was only 5 lb. 9 oz. There was marked obstruction to the airway, with dyspnoea and intercostal and neck recession; this increased if the infant was removed from the oxygen tent, when cyanosis ensued. The site of obstruction appeared to be in the nasal passages.

Examination showed blood-stained mucus in the right nostril, and a smooth, whitish tumour descending half-inch below the soft palate and causing complete nasal obstruction. Operation was decided upon because the child did not appear likely to survive otherwise.

Achromycin 100 mgm. 8-hourly orally was commenced. On the evening of 5.4.57, general anæsthesia was induced with cyclopropane and oxygen. It was very difficult to maintain an airway because the soft palate was depressed against the base of the tongue; as soon as the infant began to lose consciousness it became cyanosed. Introduction of an infant-sized airway did not help much, and it was not until a Boyle-Davis gag had been inserted that the airway became adequate. It was not considered necessary to intubate. The tumour was avulsed from its broad attachment with Luc's forceps, and came away in one piece. Brisk but transient hæmorrhage was easily controlled by suction.

Post-operative recovery was uneventful. Breast feeding was established within 24 hours. The chest deformity had disappeared by the second day. When last seen some 10 months later, the baby was symptom-free and making normal progress.

Pathology. The macroscopic appearance is shown in Fig. 2. The microscopic appearance was reported on as follows (Fig. 3):

"Sections of the tumour show it to be composed mostly of a mass of undifferentiated mesenchymal tissue, with a covering of stratified squamous epithelium. Within the mass there are scattered islands of squamous epithelium, small groups of duct-like spaces lined by cuboidal epithelium, and occasional bundles of smooth muscle fibres. There is no evidence of malignancy. As elements derived from only two of the three germ layers are seen, the tumour should be regarded as a complex hamartoma."

Case II. This was a full-term female child born on 2.11.57, the first of twins. The second twin was a male, and completely normal. There was one previous child, aged 2 years. The pregnancy was normal; both twins were easy breech deliveries. The birth-weight of this twin was 6 lb. 14 oz. It breathed immediately, and muscular tone was good, but it had considerable difficulty in maintaining an airway. There was great respiratory effort, with intercostal and neck recession and cyanosis. The obstruction to breathing seemed to be situated in the nasal passages.

The infant was seen on 3.11.57 by Mr. Lewis, and oral examination showed the tumour barely descending below the soft palate. A lateral

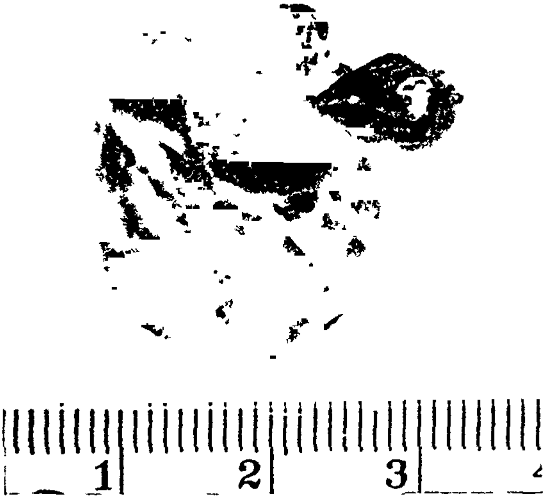

FIG 2
Case I × 2 actual size

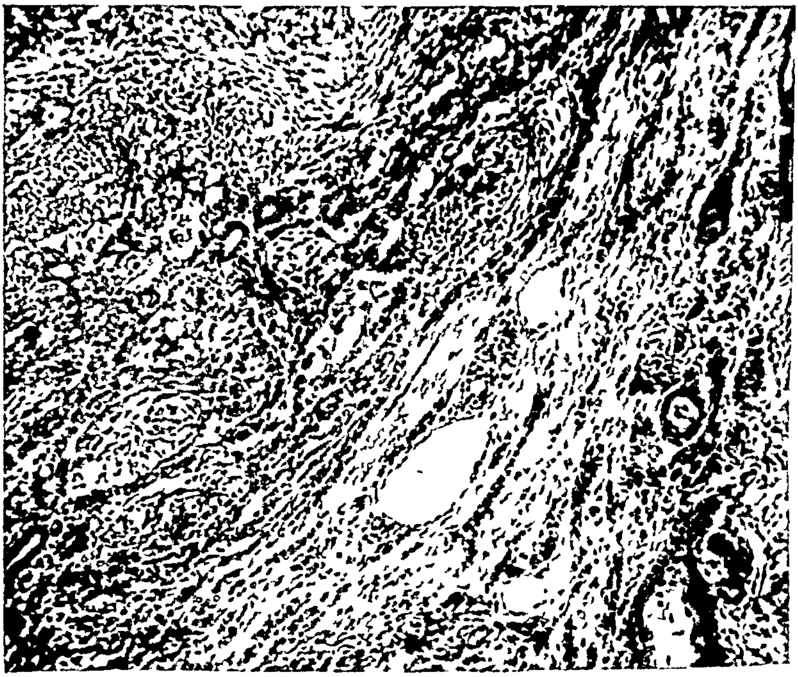

FIG 3
Case I. Microphotograph See text for description.

X-ray of the nasopharynx was taken (Fig. 1), which showed the tumour completely obstructing the nasopharynx and depressing the soft palate on to the base of the tongue. As the pink colour of the child could only be maintained in an oxygen tent, it was felt that an attempt at removal was imperative, although it was realized that removal of the bony mass might present considerable difficulty.

Crystamycin 50 mgm. 8-hourly was given systemically. On 3.11.57 general anæsthesia was induced with cyclopropane and oxygen. An endotracheal tube was passed without difficulty. The tumour was seized with grasping forceps, but on two occasions they slipped off the bony centre of the mass removing soft tissue only. On the third occasion the forceps held and the bone, together with the rest of the soft tissue, was gently rocked free from its attachment to the basisphenoid. Brisk initial hæmorrhage quickly stopped.

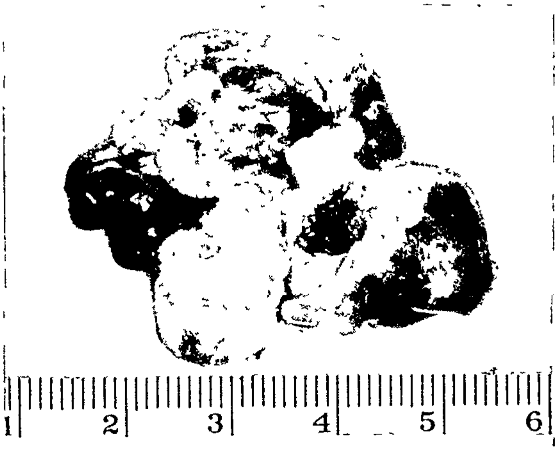

Fig 4

Case II × 2 actual size

After removal of the tumour, the infant's breathing was dramatically improved. A good pink colour was maintained, and intercostal recession stopped immediately.

When last seen 2½ months later, the baby was making normal progress.

Pathology. The macroscopic appearance is shown in Fig. 4. The microscopic appearance was reported on as follows:

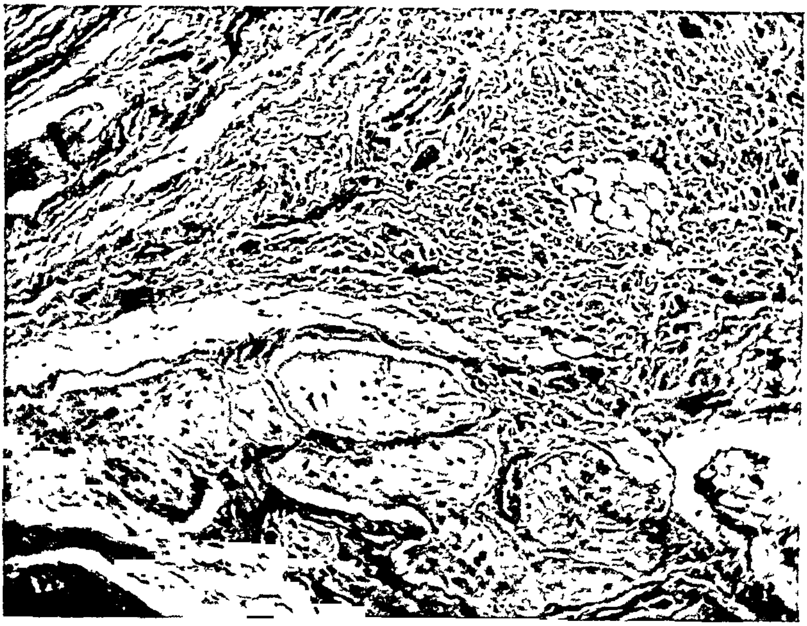

FIG. 5.
Case II. Microphotograph. Showing striated muscle, adipose, and brain tissue

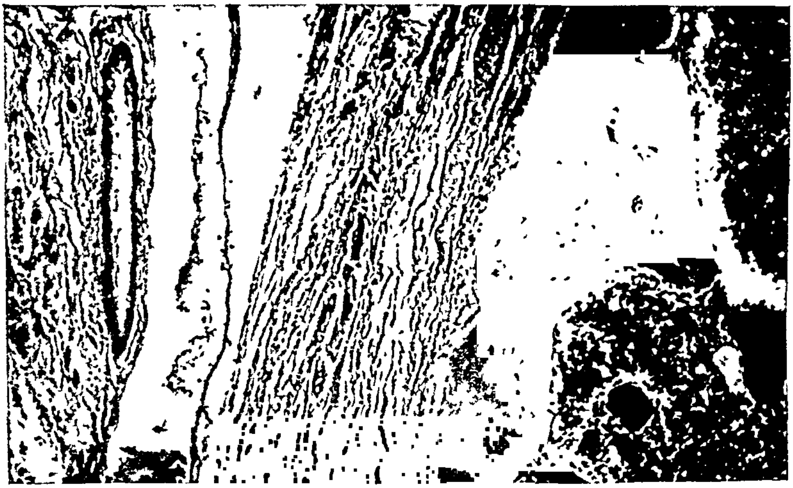

FIG 6
Case II. Microphotograph Showing bone with actively hæmopoietic marrow, connective tissue, and cyst lined by respiratory epithelium

"Sections of the tumour show it to be composed of well-formed tissues derived from the three germ layers, including smooth and striated muscle tissue, adipose tissue, and brain tissue (Fig. 5). There is an area of bone including actively hæmopoietic marrow, and very occasional minute cysts lined by respiratory-type epithelium (Fig. 6). The histological appearance is that of a benign teratoma."

Summary

The literature relating to teratoid tumours of the nasopharynx is briefly reviewed, and the pathology, clinical picture and treatment of these tumours discussed. Two cases which have occurred recently at King's College Hospital are added to the literature.

Acknowledgments

We wish to thank Mr. R. S. Lewis for permission to publish the two cases, and for his advice in the preparation of this paper. We also wish to thank Dr. M. E. A. Powell of the Pathology Department, King's College Hospital, for the reports on the sections.

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CLINICAL EXPERIENCE WITH DETERGENT NOSE DROPS AND AN EXPERIMENTAL INVESTIGATION OF THEIR EFFECT ON CILIATED MUCOUS EPITHELIUM

By P. E. ROLAND and P. G. WRIGHT* (Kampala, Uganda)

PART I

NASAL catarrh is an almost universal complaint. By the time patients see a specialist they have as a rule tried nose drops containing a variety of vaso-constrictor drugs without relief of symptoms. When examined many of these cases show no sign of infection, but are mild cases of rhinitis sicca without crusting, though the mucous membrane appears to be drier than normal. A simple alkaline nasal douche keeps most of these patients comfortable, but many dislike this somewhat messy form of treatment and prefer nasal drops. Some two years ago one of us (P.E.R.) started to try the effect of "Alevaire" (a proprietary preparation made by Messrs. Bayer Ltd.) in the form of drops or spray in this type of case, hoping that its detergent action would lead to a thinning of any mucus that was present, and its surface spreading action to a more even covering of the mucous membrane with mucus. The first results showed so much subjective improvement that a larger trial was decided on, and the makers provided a generous supply of Alevaire and its solvent in differently marked spray bottles. These were used in about 50 patients. Over 75 per cent. of the above type of case showed marked relief after Alevaire whilst about 50 per cent. showed relief after the solvent alone. The high percentage of improvement when the solvent only was used is probably not only due to psychological reasons, but also to the mucolytic effect of the alkaline solvent.

Apart from cases with mild rhinitis sicca, I have used Alevaire nose drops in cases of severe atrophic rhinitis in addition to nasal douches when they help to dislodge the crusts. Similarly I have found them beneficial in reducing post-operative crusting after nasal operations.

Though I found Alevaire effective I did not like to use it more extensively until I was certain of its effect on ciliary activity and I have not been able to find any literature where this point was critically examined The fact that it had been used in pulmonary disease for years without causing ill effects did not completely allay my fears as it was used in a much finer spray and probably reached the mucous membrane in smaller concentration In addition, Proetz (1953) found that three other detergents in a similar concentration had a deleterious effect on ciliated mucous epithelium and on ciliary activity This point has now been investigated in the Physiology Department of Makerere College Medical School

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PART II

The main function of the ciliated mucous epithelium in the respiratory tract is the transport away of foreign particles which arrive upon its surface To this end two factors contribute There is the continuous secretion of a thin film of mucus and the movement of this film by ciliary action In considering the effect of chemical substances upon this system both factors must be considered because they may vary independently and, according to the technique, affect the conclusions to be drawn from the observations

Methods

Ciliary activity was directly observed through a binocular dissecting microscope (× 80 magnification) using reflected light

Experiments were carried out upon isolated human adenoid removed at operation and upon guinea-pig and rat tracheae removed immediately after killing the animals by a blow upon the head Some experiments were also made on cat tracheae removed under sodium pentobarbitone anæsthesia On removal all tissues were at once placed in chilled oxygenated Locke's or Kreb s solution, and they were subsequently examined in the same solution at room temperature (about 23° C) or at 37° C

Ciliary transport The functioning of the transport system as a whole was assessed on isolated guinea pig, rat and cat tracheae, and upon dog tracheae *in situ* with the circulation and nerve supply substantially intact.

Sodium pentobarbitone (Nembutal) anæsthesia (50 mg/kg body weight) was maintained throughout the *in vivo* experiments

The isolated trachea was cut down the dorsal side and pinned out flat upon a cork board In order to minimize errors of measurement due to drainage artefacts the board was mounted in a closed glass dish inclined at an angle of 20° from the horizontal so that ciliary action worked against the direction of drainage A fine jet of oxygen bubbling through Locke's solution (or 95 per cent oxygen, 5 per cent carbon dioxide for Kreb's solution) in the lower angle, kept the humidity high within the dish

In some experiments in order to avoid drainage or counter-current effects the preparation was mounted horizontally and submerged beneath a few mm. of fluid.

The dog trachea was approached from the ventral surface and cannulated with a right-angled cannula as it emerged from the chest. A strip about a ¼ in. in width was cut from the ventral side of the trachea from above the cannula up to the larynx and when the bleeding from the cut edges had ceased the trachea was closed by a strip of perspex. The neck was immobilized so that the trachea was inclined at an angle of 20° from the horizontal and gravitational drainage was towards the lung end. A light was shone from above and the mucosa examined with the binocular microscope.

In both types of experiment fine carbon particles (about 20μ diameter) were sprayed upon the mucosa, or added to the fluid above it, and the time measured for selected particles to traverse a distance fixed by parallel wires in one of the microscope eyepieces. This was usually 10 mm. measured on the mucosa. Fresh particles were added before each set of measurements. In all experiments following the application of fluid one minute was allowed before measurements were begun. Ten or twelve particles were timed for each set of measurements, and each of these was representative of many more particles in the same region. Particles which were not moving well were ignored.

In the "drained" experiments the bathing solutions were applied to the mucosa by means of a spray or as small drops from a teat pipette. Excess drained off at the lower end and was removed by suction.

In all experiments the temperature of the bathing solutions was carefully adjusted before adding them to the preparation.

"Alevaire" is a solution of 0·125 per cent. oxyethylated tertiary octylphenol-formaldehyde polymer (Superinone or Triton W.R., 1339), 5 per cent. glycerol, 2 per cent. sodium bicarbonate in water and was supplied by Messrs. Bayer Ltd. It has a pH of about 9·7.

Results

Ciliary activity. Alevaire when applied directly to the epithelium produced no observable change in ciliary activity. In five experiments human adenoid tissue was placed in a moist chamber at room temperature and Alevaire was applied four times within one hour without observable effect. The tissue was then rinsed with Locke's solution and placed in a refrigerator at 4° C. It was examined at intervals after warming to room temperature and the cilia were observed to be beating effectively 50 hours from the commencement of the experiment. The tissue did not behave differently from tissue treated only with Locke's solution and handled in parallel as a control. Similarly Alevaire had no toxic action upon the ciliated epithelium of guinea-pig and rat trachea in a dozen experiments at room temperature or 37° C

Ciliary transport

In vitro Under the experimental conditions carbon dust can be seen to be carried away in a relatively smooth and continuous fashion Particles of dust remain fairly discreet and while there are variations in the rate of movement from place to place, there is a considerable degree of uniformity about the whole system This is illustrated in the first part of Fig 1 which shows the mean rate of transport and the 5 per cent confidence limits calculated for each set of measurements over a period of an hour of observation in a typical experiment When Alevaire is applied to the mucosa there is initially vigorous mixing activity, doubtless due to its surface active properties, and then the picture settles down again But it is an altered picture Dust particles now tend to be more clumped together and in many regions remain stationary The relatively homogeneous moving film is broken up There are strings of mucus carrying particles Many particles can be observed in agitation, but not in onward movement It appears as if many of the particles are no longer supported by the mucus film, but have sunk through it and are sitting directly upon the cilia

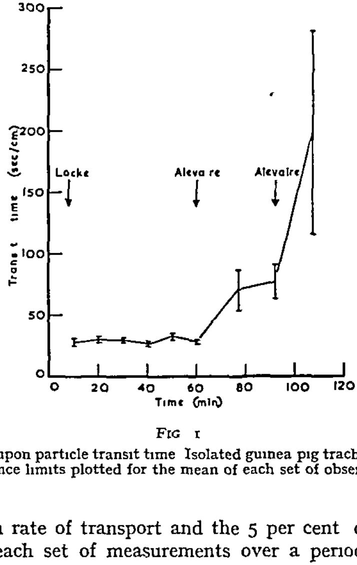

FIG 1

Effect of detergent upon particle transit time Isolated guinea pig trachea 37° C 5 per cent confidence limits plotted for the mean of each set of observations

Fig. 1 shows the effect of the application of Alevaire in undiluted form. The mean time of transport over the field distance is raised and the scatter of observations is considerably wider. However the graph cannot show the whole picture because it only takes account of moving particles. In the untreated state the vast majority of particles are being moved along, but after treatment with the detergent only a minority are moving. The majority of particles are stationary (though agitated) and do not move along during the several hours the experiment continues.

In vivo. Initially, as with the isolated mucosa there is a smooth and continuing transport of carbon dust up the trachea. When Alevaire is applied there is vigorous mixing and the uniform mucus sheet is broken up. Rafts of dust and mucus float rapidly down the trachea towards the lungs. Some particles continue their upward movement whilst adjacent ones move downwards. But unlike the isolated tissue experiments this is a temporary phase. The mucus film is re-established and orderly transport returns. Fig. 2 shows that the movement of particles is slowed and the scatter of observations is considerably increased by Alevaire but that over successive five minute periods the system returns towards its original state. A second application produces similar results.

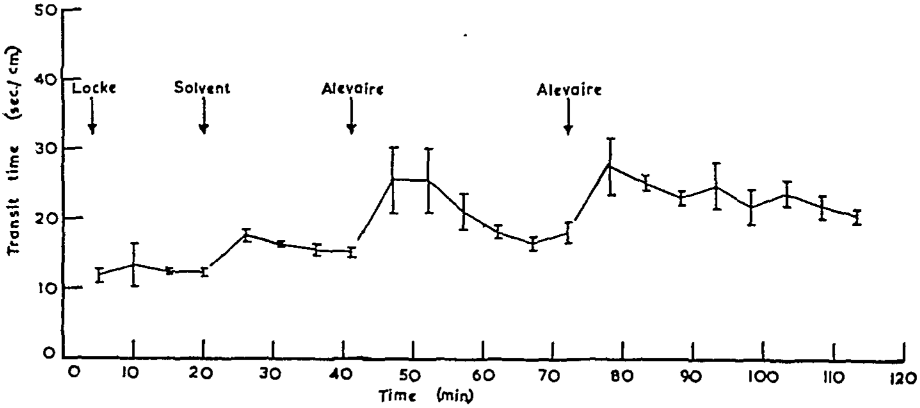

FIG. 2.

Effect of solvent and detergent upon particle transit time. Dog trachea *in situ*. 37° C. Sodium pentobarbitone anæsthesia. Five per cent. confidence limits plotted for the mean of each set of observations.

The rate of ciliary transport is several times faster in the dog tracheae *in vivo* than in isolated tracheae at 37° C. This may be due, at least in part, to the secretion of mucus which continues in the former case but not in the latter.

A solution of 5 per cent. glycerol and 2 per cent. bicarbonate—Alevaire without its detergent component—produces an overall slowing which is slight but significant (Fig 2) This figure also shows that the effect is uniform as the scatter of observations remains small and in the isolated tissue experiments it was noticeable that particles did not "stick' to the cilia as when the detergent was present

Discussion and Conclusions

Under the experimental conditions using ciliated mucous membranes, it has been shown that Alevaire in undiluted form has no detectably deleterious action upon the survival of the ciliated cells It was not possible to assess ciliary activity quantitatively The cilia were observed directly through a microscope and small changes in their rate of movement as a result of adding a solution or over a period of time would be quite impossible to detect However, gross changes would have been readily detected Since these isolated conditions would tend to mitigate against tissue survival, we feel sure that with occasional use the preparation is unlikely to harm intact mucous membranes *in situ* Proetz (1953) found that three other surface active agents in 0 1 per cent solution caused disintegration of the epithelial surface at once in isolated tissue and stopped ciliary motility in three minutes in the living rabbit

The question of the action of Alevaire upon ciliary transport of material is more difficult because it involves besides the cilia both the secretion of mucus and the condition of the mucus film The type of experiment used to assess the effectiveness of the transport system has been in use for a considerable time (Lommel, 1908, Henderson and Taylor, 1910, L Hill, 1928, Lucas and Douglas, 1934, Barclay and Franklin, 1937, Stewart, 1948, Kordik *et al*, 1952) Its deficiencies as a quantitative method, however, had not been adequately discussed until just recently (J R Hill, 1957) in a paper which appeared whilst this paper was in preparation. The necessary attention to technical detail needed for consistent results has been suitably stressed by this author and will not be reviewed here except for one point In order more nearly to simulate the conditions under which nose drops are used the drained type of preparation has been used in the majority of experiments in preference to the completely immersed type A smaller number of immersed preparations, however, have not shown different results For the reasons given by Hill the 5 per cent confidence limits for each set of measurements has been calculated (from the standard error and the t value for the appropriate number of degrees of freedom) and plotted in the figures in this paper

Our experiments with isolated tissues have shown that the surface active properties of Alevaire considerably reduce the effectiveness of the system (Fig 1) But under these conditions mucus secretion must be very small, if not completely stopped (Kordik *et al*, 1952 Dalhamn, 1956)

The effect upon the system of continuing mucus secretion is well shown by comparison of Fig. 1 with Fig. 2. The action of the detergent was similar in both cases but in the latter case, *in vivo*, the effect was short lived and the system returned towards its original state of effectiveness. The importance of the mucus film in the removal of foreign bodies from the mucosa is perhaps not often appreciated, Florey *et al.* (1932) said, "the ability of the tracheal mucosa to remove small foreign bodies depends to a certain extent on the 'stickiness' of the mucus". When this stickiness is impaired by a surface active substance then the effectiveness of the system is also impaired. Florey *et al.*, also found that when mucus secretion was stimulated foreign particles moved more rapidly up the trachea. Our experiments did not suggest that the preparation stimulated mucus secretion, but any reflex secretion might well be suppressed by the anæsthesia. Lucas and Douglas (1935) also suggested that changes in the rate of particle movement produced by drugs were due to changes in the state of the mucus and in its rate of secretion.

The observations of Lucas and Douglas (1934) suggested that "the mucus sheet is composed of two layers—an outer stratum of mucus, which rests on the tips of the moving cilia, and an inner fluid layer of low viscosity, which forms a suitable medium for the vibrating cilia". Our observation that after the application of the surface active substance foreign particles no longer appeared to be supported in the mucus film, they appeared to be sticking to the cilia, to be agitated, but not moved onwards, would accord with this theory. The mucus sheet was broken up and the conveyor belt ceased to function, though the individual moving parts were unaffected.

However, when the epithelium or the mucus is in an abnormal state other factors must be considered. Thick viscid mucus delays the cleansing action of the system (L. Hill, 1928; Tremble, 1947). Viscid muco-pus will probably be broken up and its removal thus facilitated by the occasional use of Alevaire. In the *in vivo* experiments the material which was dislodged from the mucosal surface by the preparation and drained down towards the lung would probably have elicited a cough reflex had this not been suppressed by anæsthesia.

If the preparation was so applied that a high local concentration was maintained it would appear unlikely to have beneficial effect. This is a possible explanation of the negative results which Holborow (1957) obtained using Alevaire in sinusitis. The preparation was placed into the maxillary antrum and was still in evidence a week later. In clinical practice such high concentrations are very rarely used.

It would appear that the main indication for the use of Alevaire is the presence of excessively sticky mucus which ciliary activity has not been able to move. By reducing the viscosity of the mucus its clearance may be facilitated even though the preparation may temporarily make the ciliary system less effective. In acute laryngo-tracheo-bronchitis it is often used continuously in oxygen tents as an aerosol (Sadove *et al*, 1954, Gans, 1954). Under these conditions it may not reach concentrations in the trachea and bronchi which would change the surface tension sufficiently to impair the transport of bronchial secretions. It should, however, be considered whether intermittent use is not preferable to a continuous one. When Alevaire is used in the upper respiratory tract in the form of drops or spray, concentrations high enough to break up the mucus film are probably obtained. Fresh secretion will soon dilute the Alevaire sufficiently to restore the normal transport of mucus. The more viscous the nasal secretions have been before the application of Alevaire, the less marked will be the deleterious effect on the ciliary system, or this may not be present at all. We, therefore, have no hesitation in continuing the use of Alevaire in all forms of rhinitis sicca. Like any other drug, it should not be used without proper indication and not to excess.

Summary

1. A proprietary preparation of alkaline detergent, called "Alevaire", in the form of drops or spray, has been found of benefit in rhinitis sicca. Its effect on ciliated mucous epithelium *in vitro* and *in vivo* has been investigated.

2. Alevaire does not impair ciliary activity.

3. Alevaire makes ciliary action less effective by altering the surface tension of the mucus. *In vivo* this effect appears to be temporary.

We wish to thank Messrs. Bayer Ltd., for the generous supply of Alevaire and its solvent used in the clinical trial and the experiments.

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CLINICAL RECORDS

HEAD INJURY WITH FRACTURE OF STAPES

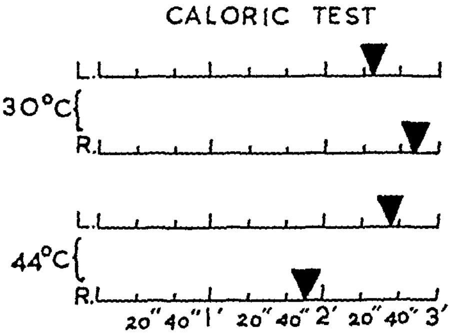
By R. A. WILLIAMS (London)

TRAUMATIC conductive deafness may be due to perforation of the tympanic membrane, to dislocation of the incus (Thorburn, 1957), or to fracture of the neck of the stapes, as in this case.

The patient was a lady, aged 35, who had been involved in a road accident on *April 1st, 1957*. She was concussed for a few minutes. There was some bleeding from the right ear. Deafness and tinnitus of the right ear were noticed immediately, and persisted until her operation. X-rays of the skull did not demonstrate any fracture. On *April 6th* she developed a right facial palsy, which was recovering ten days later.

On examination, the left tympanic membrane was normal. The right tympanic membrane was also normal except for a dark mark postero-superiorly looking like a blood clot in the middle ear. Weber's test was referred to the right, and Rinne's test was negative on the right side, positive on the left. Absolute bone conduction was normal on both sides. The right eustachian tube was catheterized and found to be clear.

The audiogram shows the degree of preoperative conductive deafness. Caloric reactions:



This shows a slight left directional preponderance, indicating some right utricular paresis.

On *October 5th* the right middle ear was explored by Mr. H. D. Fairman. . . .

stapes. The denuded part of the external meatus was grafted. This was, in effect, a Wullstein tympanoplasty Type 3, the difference being that there was no mastoidectomy. For a few days after the operation the patient complained of a sweet taste on the right side of the tongue.

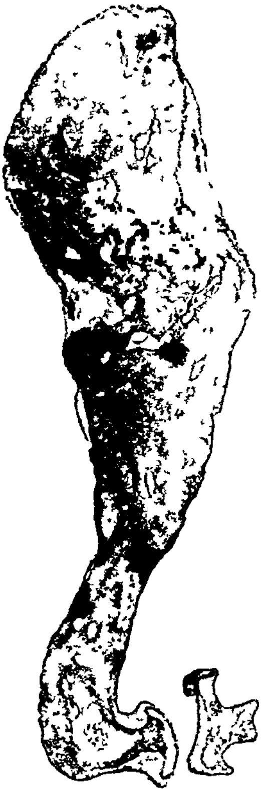
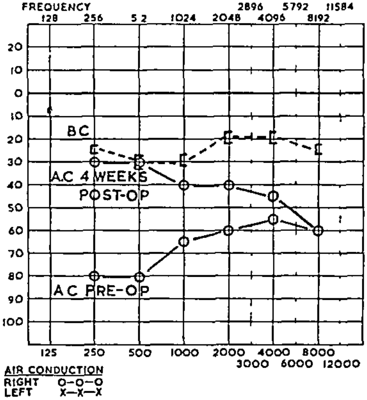


FIG 2

Section through incus, and head and neck of stapes

The photograph and sections show the fracture site in relation to the incudo-stapedial joint, and that squamous epithelium had grown over the fractured surface of the bone. This shows that there was permanent non-union of the fracture. The audiogram shows the degree of hearing improvement four weeks after operation. It is possible that there was also some inner-ear damage as suggested by the caloric tests, and that this had contributed to the perceptive element of the deafness.



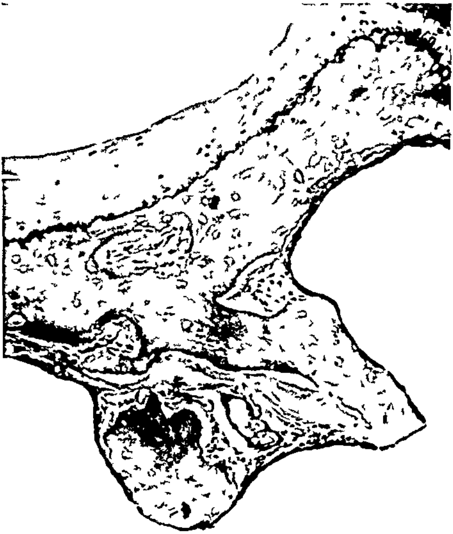


Fig 3

High power view of head and neck of stapes showing fracture surface epithelized

Acknowledgments

This patient was under the care of Mr. H. D. Fairman, and my thanks are due to him for permission to publish his case. I would also like to thank Dr. Sandry, and the technicians of the pathology and photographic departments of Frenchay Hospital for preparing the slides and photographs.

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A SIMPLE NASOPHARYNGEAL TERATOMA

By C A HOLBOROW (London)

A FURTHER case of neonatal asphyxia due to a simple naso-pharyngeal teratoma or "hairy nasal polyp" is recorded

Case Report

F R , the first child and the daughter of a doctor, was born at full term by normal delivery The birth weight was 7 lb 7 oz Immediately after delivery there was considerable respiratory distress and cyanosis with evidence of intermittent laryngeal obstruction Examination showed a polypoid tumour hanging down from the post nasal space and obstructing the airway No other abnormality was found A Portex endotracheal tube was passed relieving the obstruction and a stomach tube was also passed to exclude œsophageal atresia This tube passed into the stomach without difficulty and some 2 c c of clear mucus was aspirated

She was transferred to The Hospital for Sick Children where further examination showed a pink, firm polyp covered with fine hair arising in the nasopharynx and descending to the level of the laryngeal inlet The polyp was avulsed without difficulty by means of a snare passed through the nose and was lifted out through the mouth No anæsthesia was used The tumour appeared to arise from the basisphenoid in the midline and there was almost no bleeding after removal Examination of the nasopharynx and larynx showed no other abnormality After this operation the infant's condition was satisfactory An X-ray of the base of the skull showed a normal appearance and she was returned to her mother at Queen Charlotte's Hospital the following day Her subsequent history has been uneventful

The polyp (Fig 1) measured 5 × 1 × 1 centimetres and consisted of fatty tissues with a small piece of cartilage covered by hair-bearing skin There was no evidence of malignancy The constricting grooves are of interest, the upper, presumably, being caused by the nasopharyngeal isthmus and the lower, possibly, by the ary-epiglottic folds

Comment

Simple nasopharyngeal teratomata are not common Kesson (1954) found records of 11 cases over a period of 20 years of which six were of successful removal of such tumours in the neonatal period He added one more Ehrich (1945) reviewed 73 cases of hairy polyp in the literature in an article on Teratoid Parasites of the mouth and nose He divided these teratomata into Epignathi, Epipalati and Episphenoids It is to this last, and most common, group that the present tumour belongs

The history is very similar to those recorded in other cases (Kesson, 1954 and Wilson, 1931). Neonatal respiratory obstruction was caused by the ball-valve action of the tumour at the laryngeal inlet. This was relieved temporarily by intubation and removal of the polyp was performed without difficulty with very little bleeding. Early recognition by the obstetrician is, of course, vital.

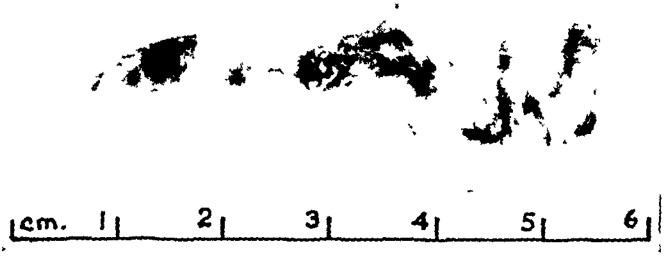

FIG. 1.
The tumour after removal.

With the simple type of teratoma obstruction does not always occur in the neonatal period and may develop some weeks or months after birth. The more highly developed teratomata are usually found in still-born infants.

Acknowledgments

I am most grateful to both Mr. James Crooks and Dr. A. White Franklin, under whom the infant was treated, for permission to publish this case.

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A CASE OF CONGENITAL PALATO-PHARYNGEAL SYMPHYSIS

By A COYAS (Athens)

Case History

Mrs P.A. aged 44 years (nun).

This patient was seen in the Ear, Nose and Throat Department of Hippokration Hospital in Athens on *15.11 56*, complaining of slight sore throat and severe difficulty in breathing through her nose, headaches, impossibility to blow her nose freely and progressive deafness for the last 15 years.

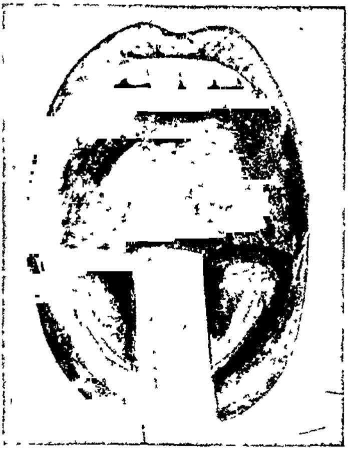

Fig. 1.

The pharynx, showing the adhesion of the soft palate to the posterior pharyngeal wall

Physical Examination

A healthy looking woman.

E.N.T. Examination

Pharynx (Fig. 1) There is just an outline of the anterior and posterior pillars, no tonsils (she had never been subjected to a tonsillectomy). The free edge of the soft palate is attached to the posterior wall of the pharynx with an exception in

FIG. 2a.

Opaque material retained in nose Very small communication between nasopharynx and pharynx

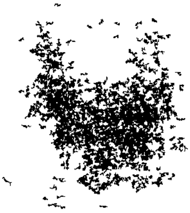

FIG. 2b.

Submento-vertical view

the middle where there is a fistula not bigger than ½ a centimeter in diameter, by which the nasopharynx communicates with the oropharynx When pressing the soft palate with a probe near the fistula, thick pus is coming out Hypopharynx normal

Nose No severe abnormality on the anatomical structure revealed under the anterior rhinoscopy, after sucking out all the mucopus from the nostrils A thin elastic catheter could pass freely through the right nostril and through the fistula The same catheter could pass with difficulty through the left side as if the choana was partially obstructed

Ears Not abnormal aspect of the tympanic membranes

Chest X-ray Nothing abnormal Blood examination Normal Wasserman Negative Kahn Negative Opthalmological examination N A D X-ray of the head with opaque substance injected in both nostrils was performed (Fig 2a and 2b)

The patient refused any operation and left the hospital one week after

The case is thought worth recording because of the unusual and very rare sort of this congenital malformation, and only very few cases have been recorded the last 50 years in the international bibliography

My thanks are due to Prof J Chryssicos under whose care this patient was, for permission to submit the case for publication

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DISLOCATION OF THE INCUS DUE TO HEAD INJURY

By FRANCIS BAUER (Liverpool)

FORMERLY, in cases of post-traumatic deafness, diagnosis of a disrupted ossicular chain, if made at all, meant that the prognosis was poor, because the condition was not considered amenable to surgical treatment. This paper describes a case diagnosed pre-operatively as a traumatic dislocation of the incus and successfully treated by surgical means.

Most cases of traumatic dislocation of the incus are due to head injuries, especially to longitudinal fracture of the petrous pyramid, which often causes paralysis of the facial nerve and deafness, each of which may or may not recover spontaneously and independently. As the facial paralysis is the more conspicuous and disabling, its conservative and operative treatment has often been discussed. Although the otological aspects as to the type of lesion (middle-ear or inner-ear deafness, vestibular function) and prognosis have been considered (Davis, 1931; Grove, 1942; Escher, 1948; Proctor *et al.*, 1956) management in the past has been only conservative. Recent advances in microsurgery of the ear, better understanding of the physiological principles underlying the function of the middle ear and antibiotic cover have made it possible to restore useful hearing by reconstruction of the middle ear and tympanic membrane, damaged by chronic suppuration. The idea of trying to improve hearing damaged by *traumatic* disruption of the ossicular chain suggested itself. Thorburn (1957) explored a case of post-traumatic middle-ear deafness and found the head of the malleus fixed by ossification inside the attic and dislocation of the incus. He mobilized the drum in order to bring it in contact with the head of the stapes, closed the upper tympanum with a skin graft and achieved a very satisfactory hearing result. Gisselsson (1958) treated a bilateral luxation of the incus by interposition of the dislocated incus between the stapes and the tympanic membrane.

The following case has been diagnosed pre-operatively as a traumatic dislocation of the incus and operated upon with the object of bringing the tympanic membrane in contact with the head of the stapes, thus producing Zoellner's "columella effect".

A 23 years old fitter was involved in a motor cycle accident on *4.10.57* in Derbyshire and suffered a fracture of the right temporal bone (Fig. 1). He was unconscious for about 36 hours and spent a week in a hospital. There was bleeding from the right ear following the accident, but no giddiness.

He was seen 9 weeks later in Walton Hospital, Liverpool, with paresis of the right facial nerve of the peripheral type and he was complaining of deafness in the right ear, which he attributed to the accident.

There was a scar in the postero-superior quadrant of the right tympanic membrane. The tympanic ring appeared intact (no notching). Rinné's test was

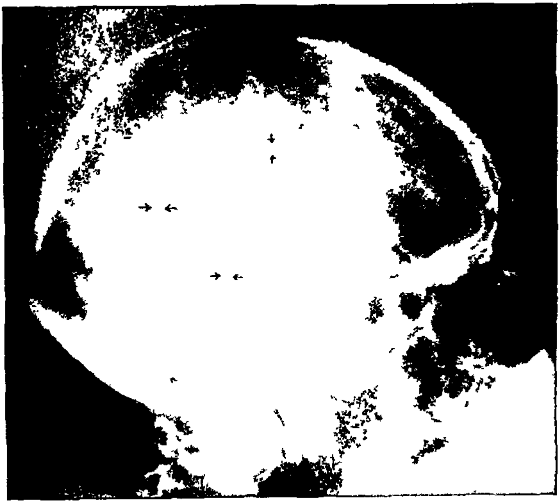

FIG 1

Fracture of the right temporal bone affecting the squama and passing down through the roof of the mastoid antrum and tympanum

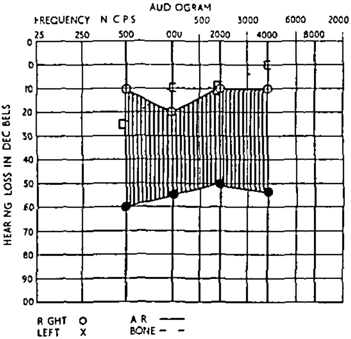

FIG 2

Audiogram showing the gain in hearing following tympano stapediopexy preoperative air conduction ○—○ post operative air conduction [[[bone conduction negative in the affected ear and on Weber's test the sound was lateralized to the right. Audiogram confirmed severe middle-ear deafness in the right ear (Fig. 2). The left ear was normal in every respect and there were no other abnormal findings.

The facial nerve recovered completely within the next six weeks, but the deafness remained unchanged. The radiological appearances of a well pneumatized mastoid supported the diagnosis of traumatic origin of the middle-ear deafness.

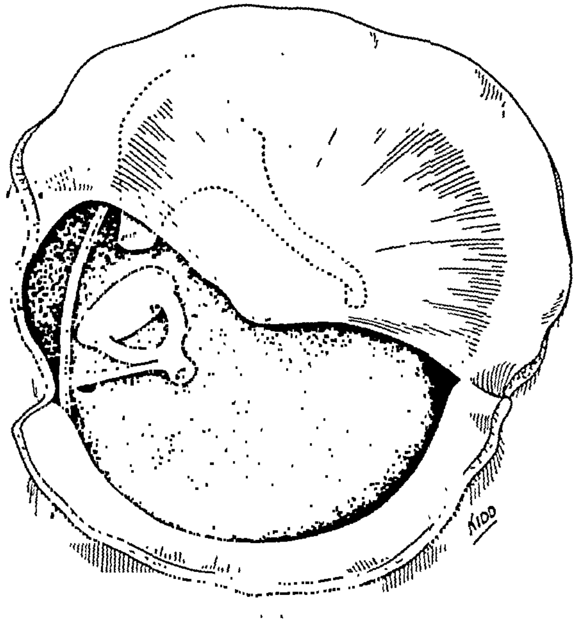

FIG. 3.

Appearances after reflection of the posterior half of the tympanic membrane forwards over the handle of the malleus (Rosen's flap). The stapes is isolated, the long crus of the incus is dislocated forwards and upwards and is lying almost horizontally above the stapes. The short process of the incus is lying in the region of the descending part of the facial nerve.

With the pre-operative diagnosis of traumatic interruption of the ossicular chain, most likely due to dislocation of the incus, on *12.2.58* the right middle ear was explored. Under general anæsthesia the auditory meatus was enlarged by an incision of the upper meatal wall from the pars flaccida of the tympanic membrane between the tragus and the crus helicis. After insertion of a small retractor, Rosen's flap, including the posterior half of the tympanic membrane and about 4 mm. of the posterior cutaneous meatal wall, was reflected forwards over the handle of the malleus. A crescent of bone of about 2 mm. width was removed from the posterior margin of the tympanic ring with Rosen's curettes and a small gouge. The head of the stapes could now be well seen, quite isolated and not articulating with the incus (Fig. 3). The stapes was mobile. The incus, however, was not immediately visible After further removal of bone from the upper margin of the tympanic ring the short process of the incus was found lying in the region of the descending part of the facial nerve, the long crus having swung forward and lying almost horizontally above the stapes It did not seem feasible to reduce the dislocated incus and hope to preserve its blood supply The incus was, therefore, removed and found intact The middle ear mucosa was normal and the Fallopian canal appeared intact It had been planned originally simply to reposition the tympano meatal flap on the head of the stapes to produce tympano stapediopexy (columella effect), but on replacement of the flap the scar in the posterior part of the tympanic membrane gave way and a perforation of the drum appeared, through which the isolated stapes was clearly visible A full-thickness graft from the postauricular skin was, therefore, taken and used for closure of the perforation as for myringoplasty Gelatine sponge was placed on top of the graft in order to maintain apposition of the tympanic membrane and of the graft to the head of the stapes and a BIPP pack placed into the meatus The pack was removed 10 days later The patient's hearing gradually improved in the ensuing six weeks On *31 3 58* the ear was dry, the perforation of the tympanic membrane had healed and audiogram revealed a hearing gain amounting to some 25-45 decibels for the speech frequencies (Fig 2) A month later no further change had occurred

Discussion

During decompression operations on the Fallopian canal for facial paralysis of traumatic origin dislocation of the incus has been noted and mentioned in passing by a number of authors (Cawthorne, 1956, Kettel, 1957, Feinmesser, 1957), but the problem of restoration of hearing was not considered Kettel even discusses the question of destroying the middle ear in the course of repair of the facial nerve, if the nerve be injured in the horizontal segment, as it may be necessary to perform a radical mastoidectomy in order to repair the nerve In one of his cases the incus was displaced into the middle ear, pushing the malleus outwards in another it was dislodged into the middle ear, which was filled with scar tissue

Cawthorne (1956) found dislocation of the incus in six out of seven cases of traumatic facial paralysis operated upon He, therefore, suggested that in cases of skull fracture with facial palsy, dislocation of the incus, as evidenced by conductive deafness and an abnormal appearance of the tympanic membrane, was a definite indication for exploration of the facial nerve in the Fallopian canal No remedial operative procedure for the relief of the conductive deafness was, however, suggested

Crabbé (1956) describes a case of longitudinal fracture of the petrous pyramid in which dislocation of the incus was diagnosed by otoscopic examination "The handle of the malleus has lost its oblique direction downwards and backwards, it hangs vertically and pointing rather forwards The pars flaccida of the tympanic membrane is scarred It is lifted up by a mass which is probably the head of the malleus detached from its handle It is luxated with the incus, which projects behind " There was a mixed type of deafness Escher (1948) describes one case of longitudinal fracture of the petrous pyramid with dislocation of the incus. There was a mixed type of deafness. Rowbotham (1949) mentions dislocation of the ossicles as a possible cause of middle-ear deafness without further details about the type of dislocation.

Longitudinal fractures of the petrous pyramid are usually extensions from fractures of the temporo-parietal region affecting the squama and passing down toward the external auditory meatus through the roof of the mastoid antrum, roof of the tympanum and along the anterior aspect of the pyramid through the superior wall of the Eustachian tube and carotid canal (Proctor *et al.*, 1956). Clinically this is the most common type (Fig. 1) because transverse fractures affecting the inner ear are usually associated with a fatal fracture of the base of the skull.

From the ossicles of the middle ear the incus is the most prone to dislocation. The handle of the malleus is attached to the tympanic membrane and its neck fixed by the anterior and posterior ligaments. The stapes is held in the oval window by the annular ligament. But the incus is attached only at its short process by ligamentous fibres to the fossa incudis, its long process with the delicate incudo-stapedial joint being unprotected and the most vulnerable link in the ossicular chain. The dislocation starts at the incudo-stapedial articulation, the long process of the incus being displaced forwards, the head of the incus rotates backwards and downwards anti-clockwise, carrying the short process of the incus downwards and forwards. The degree of dislocation varies from luxation in the incudo-stapedial joint (Thorburn's and Gisselsson's cases) to total dislocation (case described) and even ejection of the incus through the tympanic membrane (Voss, 1936).

In cases of longitudinal fracture of the petrous pyramid permanent deafness of the pure middle-ear type is much rarer than a mixed type of deafness because of some coexisting involvement of the inner ear or acoustic nerve. It is difficult to give any definite figures because since the introduction of audiometric tests the findings of an inner-ear component in traumatic deafness have increased. Older authors give a much higher proportion of pure conduction deafness. The explanation for this discrepancy lies in the technical difficulty of establishing the upper limit of hearing acuity with tuning forks of high frequency.

As dislocation of the incus is now amenable to surgical treatment, the correct diagnosis of this condition assumes practical importance. There is no doubt that in the past the possibility of this dislocation has either not been considered and post-traumatic middle-ear deafness attributed to other, probably non-existing conditions, e.g. adhesions from organizing hæmatoma, fracture of the ossicles, ankylosis of the stapes more or less complete (Piaget *et al.*), or, if considered, the diagnosis has been rejected on insufficient grounds. Thus Crabbé *et al.* found in cases of longitudinal fracture of the petrous pyramid that conduction deafness persisted in a number of subjects, although clinical examination could not demonstrate a lesion of the ossicular apparatus, "the tympanic membrane being healed and the handle of the malleus moving normally on testing with Siegle's speculum." This test certainly does not exclude dislocation of the incus, as the handle of the malleus does move whether or not the incus is dislocated. For conclusive evidence of the dislocation the tympanic membrane must be reflected and the incudo-stapedial articulation inspected directly.

The diagnosis of dislocation of the incus should be considered in all cases of permanent middle ear deafness or mixed type of deafness with a marked middle ear component, as evidenced by good bone conduction, following head injury. Further physical signs may be present notching or irregularity of the bony tympanic ring, scarring of the tympanic membrane, facial paralysis of the peripheral type associated with a longitudinal fracture of the petrous pyramid The presence of good pneumatization of the mastoid process is a further supporting evidence that the middle-ear deafness is more likely traumatic than inflammatory in origin

Most authors recommend exploration of the facial nerve if no sign of recovery occurs within two months of injury, because intraneural fibrosis is likely to interfere with regeneration of the nerve Similar considerations do not hold for reconstruction of the middle ear, following dislocation of the incus, because mobility of the stapes remains unimpaired for many years (in Thorburn's case 15 years, in Gisselsson's case 8 years) Thus there must be a number of cases of traumatic middle-ear deafness still amenable to surgical treatment even many years after the accident Apart from the purely medical point of view, the medico-legal aspects are of interest in cases of compensation

Hitherto only persisting facial paralysis following head injury was considered an indication for exploration of the middle ear The case described shows that persistent middle-ear deafness, in which dislocation of the incus is diagnosed, should also be deliberately explored with view to restoring useful hearing by reconstruction of the middle ear Traumatic middle ear deafness should now be considered an indication for surgical treatment

Summary

Permanent middle ear deafness following head injury has been attributed in the past to fracture of the ossicles, adhesions from organizing hæmatoma and ankylosis of the stapes and treated expectantly The real cause of post-traumatic middle-ear deafness is dislocation of the incus The signs and symptoms of dislocation of the incus are described and a case reported, in which the diagnosis was made pre-operatively Tympano-stapediopexy restored useful hearing The diagnosis of dislocation of the incus should be kept in mind in cases of post-traumatic middle-ear deafness and if made, surgical treatment by reconstruction of the middle-ear is recommended

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SOCIETIES' PROCEEDINGS

ROYAL SOCIETY OF MEDICINE—SECTION OF OTOLOGY

President: I. SIMSON HALL, F.R.C.P.Ed., F.R.C.S.Ed.

February 7th, 1958

Discussion on Cholesteatosis of the Attic

THE opening papers will be found on pp. 593-625.

Discussion

MR. F. MCGUCKIN said that he was unable to agree with the view expressed by Professor Rüedi that erosion of bone was caused by pressure of cholesteatoma. This was certainly not the whole explanation because there were too many exceptions, and it might not be a factor at all. He also thought that under certain circumstances in-driving of the flaccid membrane in negative pressure deafness could set up cholesteatoma. The introduction of moisture or of oil might set the process going and he thought that such a process could be observed in adults. Moreover there might possibly be some relation between cholesterol granuloma and cholesteatoma, but not in the fashion suggested some years ago by Mr. R. R. Simpson. Black cholesterol granuloma in the mastoid could perhaps be secondary to cholesteatomatous obstruction in the attic. It was something new to find that Mr. Tumarkin now held hypocellularity to be not invariable in the presence of chronic otitis media and it was pleasing to note that he had abandoned his famous atticotomy, which was purely a drainage procedure, in favour of excisive surgery—still leaving room for separate consideration of function. The term "retracted drum" should be avoided, since it did not express the true mechanism. The alteration in the contour of the drum head arose from difference in pressure so that it was in-driven and not retracted. It seemed a pity that a highly logical person should indulge in a term which savoured of slack thinking. Nevertheless the contributions of all three speakers had provided magnificent instruction. Mr. Birrell had stressed that inadequate medical treatment in acute ear disease led to trouble. When we employ medical treatment we must tighten our canons of recovery, and not hide behind the word masking.

MR. I. B. THORBURN said that he was concerned about the surgical management of attic cholesteatoma. He showed colour stills from three cases in which the spread of cholesteatoma from the attic towards the antrum and the upper tympanum was successfully contained by an expansion of mucous membrane.

He considered that under the operating microscope it was possible to dissect out the cholesteatoma completely whilst retaining the protective layer of mucous membrane. In some such cases the tympanum had sealed itself off from the disease and had established a natural type II or III tympanoplasty. In others, especially with ossicular caries, it was necessary to complete a tympanoplasty operation using full thickness skin graft enclosing the region which had contained cholesteatoma with the assurance that the disease had been completely removed.

Mr. R. R. Simpson congratulated Professor Rüedi on the masterly survey of the many conflicting views on the origin of cholesteatoma, and on the presentation of the recent work on the subject. Professor Rüedi's paper will be read in future with the great interest that its importance deserves. The view of bone absorption by pressure, though still held, was suspect and should be re-examined.

It was always interesting to hear Mr. Tumarkin's presentation of his new spring collection of his views on this subject, but the suggested new terms merely befogged the issue still further. Mr. Simpson supported Rüedi and Friedmann in their plea that the term cholesteatoma should be retained, and modified when necessary by a definite adjective.

GENERAL NOTES

The Editors are anxious to improve the clinical aspect of the Journal and would welcome more reports of interesting cases These clinical records need not be at all elaborate and it would be unnecessary to go minutely into the ætiology or the literature of the subject Senior Registrars have unique opportunities for seeing a large variety of cases, many with unusual features It is in many ways regrettable that these should go unrecorded It is hoped that all those in charge of clinics will help us in this matter and see that such cases are not overlooked

THE BÁRÁNY JUBILEE MEDAL

In conjunction with the graduation ceremonies at the Uppsala University, Sweden, the Bárány Jubilee Medal was awarded to Dr Charles Skinner Hallpike, M D , F R C P., F R C S , F R S , Medical Research Council, Otological Research Unit, National Hospital, London

At the ceremony Dr. C O Nylén, the successor of Bárany in the chair of Otology at Uppsala University gave an outline of Hallpike's works

The Medical Faculty of Uppsala has awarded this outstanding distinction for 1958 to the eminent and internationally known scientist, Dr C S Hallpike, London *By his clinical and experimental investigations he has increased to* a high degree our knowledge of the functions of the vestibular system and of optomotorial nystagmus

The Dean of the Medical Faculty then handed the gold medal to Dr Hallpike

THE INSTITUTE OF LARYNGOLOGY AND OTOLOGY

330-332 Gray's Inn Road, London, W C 1

The Academic Board announces the following

SPECIAL LECTURES AND DEMONSTRATIONS

to be given at the Institute—except where otherwise stated These are open to all members of the Specialty and to postgraduate students No ticket of admission is required

Friday, 8th August, 1958, at 5 30 p m (instead of 30th July as previously notified)

Dr Ben H Senturia (St Louis)—"Diseases of the external ear "

Friday, 24th October, 1958, at 5.30 p.m.
PROFESSOR C. V. HARRISON (Postgraduate Medical School of London)—"The pathology of malignant lymphomas."

Saturday, 8th November, 1958, at 11.30 a.m.
PROFESSOR LUCIUS RÜEDI (Zurich)—"New developments in the Surgery of Oto-sclerotic Diseases."

Friday, 14th November, 1958, at 4.30 p.m.
MR. T. G. WILSON (Dublin)—ANNUAL ADDRESS.

Friday, 12th December, 1958, at 5.30 p.m.
PROFESSOR W. ST. C. SYMMERS (Charing Cross Hospital Medical School)—"Mycotic diseases".

PROVISIONAL LIST OF SHORT COURSES FOR CONSULTANTS AND SENIOR REGISTRARS

3rd and 4th October, 1958—Practical Aural Surgery Class.

17th, 18th and 19th February, 1959—Malignant diseases of the ear, nose and throat.

21st and 22nd February, 1959—Pathology of the ear, nose and throat.

10th, 11th and 12th March, 1959—Diagnosis and treatment of the deaf child.

13th and 14th March, 1959—Modern methods of acoustic and vestibular testing.

10th and 11th April, 1959—Practical Aural Surgery Class.

The number of entries is limited in each course.

A detailed syllabus of any of the above can be obtained on application to the Secretary.

SEMON LECTURE

THIS lecture will be delivered by Professor Luzius Rüedi of Zürich in the Barnes Hall of the Royal Society of Medicine on Thursday, 6th November. Subject : Some observations on the Histology and Functions of the Larynx.

The Journal of Laryngology and Otology

(*Founded in* 1887 *by* Morell Mackenzie *and* Norris Wolfenden)

September 1958

EDITORIAL

Journal of Laryngology and Otology

SINCE 1920 the subscription rate of the *Journal of Laryngology and Otology* has remained constant, except for one change, when in 1950 the price was raised from £2 to £3 3s od

Owing to the increased cost of printing and postages a further increase is unavoidable As from January 1st, 1959, the subscription rate of the Journal will be £4 4s. od , including postage

Seventeen-Year Index

A seventeen-year Index covering the years 1940 to 1957 is in course of preparation, and will be published early in 1959 A copy of this volume will be sent free to every subscriber It is hoped that this will be found a valuable aid for reference

If any subscribers find that they are short of back numbers, a certain number of these are available on application to the publishers, Messrs Headley Brothers Ltd , 109 Kingsway, London, W C 2.

FURTHER CONTRIBUTIONS TO THE DEVELOPMENT OF THE LABYRINTHINE CAPSULE

By A. ŠERCER and J. KRMPOTIĆ (Zagreb)

Introduction

In a special study concerning the relations of otosclerosis to anthropology one of us tried to prove that the labyrinthine capsule repeats in the course of its ontogenetic development the principal stages of phylogenesis. In order to enlighten this problem we must point out the difference between the human labyrinth and that of other vertebrates. This difference can be easily seen in Fig. 1 drawn according to the statement of Delattre. This drawing shows us that the lateral canal keeps in both cases its horizontal position. This canal plays the part of a stabilizer of the skull in space. The posterior canal on the contrary changes its position in man; it is shifted downwards riding thus over the lateral canal. The round window is situated in other vertebrates lateral to the oval window, while in man it lies below it following the displacement of the cochlea forwards and upwards.

This process of phylogenetic transformation of the labyrinthine capsule can be followed on a series of schematic drawings (Fig. 2). There we can see that the shape of the labyrinthine block changes in such a way that the cochlea, which occupies the inferior square, is shifted several millimetres forward; this facilitates the rotation of the upper square backwards and downwards. Finally the lower block is rotated around its transverse axis forwards and upwards thus giving the labyrinth its definite shape.

This marked difference in the shape of human and vertebrate labyrinths has been mentioned and described by many anatomists and anthropologists but Delattre was the first to express the idea that this difference is the result of the erect posture of man. Studying the development of the skull for years, Delattre has contributed much to the knowledge of the phylogenetic development of the skull by his ingenious investigations concerning the rotation of the pyramids and occiput. He propounded, already in 1952, the question whether the human labyrinth repeats in its ontogenetic development the stages of phylogenesis.

This problem was of special interest for us because we hoped to find in the development of the labyrinth the solution of certain pathologic processes.

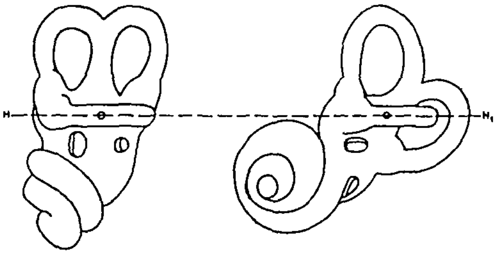

Fig 1

The labyrinth of carnivora and man The vertical canals of the former lie above the lateral semicircular canals while in man the posterior canal rides over the lateral one The cochlea is situated in carnivora below the lateral semicircular canal while in man it lies in front of it Fenestra rotunda is in the first case situated behind the oval window and in the second case below it $H\ H_1$ represents the ortovestibular line which runs horizontally

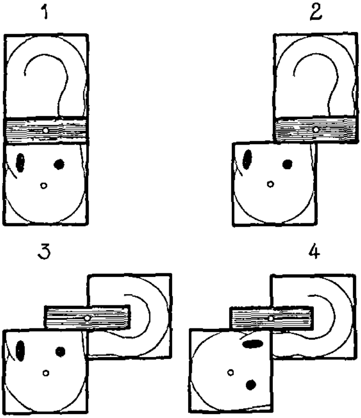

Fig 2

The geometrical analysis of the phylogenetic development of the labyrinthine block (1) The block of vertical canals is situated above the cochlear block By a dotted line is represented the block of the lateral canal The longitudinal diameter of the oval window is perpendicular and the round window is situated behind the oval one (2) The cochlear block is gliding forwards (3) The upper block can thus rotate through 90° so that the posterior canal rides over the lateral one (4) The lower block has been rotated too for 90° so that the round window is coming to lie below the oval window the longitudinal axis of which lies now parallel with the horizontal canal The block of the lateral canal never changes its position it remains always horizontal

The human labyrinth has actually, in early stages of embryonic life, a shape that reminds one very much of the labyrinth of other vertebrates. This can be seen clearly if we compare the human labyrinth of a foetus of three months with a carnivorous labyrinth. This fact has been already proved by the famous work of W. His (1889) and Hyrtl (1845). The ontogenetic development of the human labyrinth, however, has not been studied as yet and that is why we tried to clear this question by our own investigations.

For this purpose we used disarticulated skulls of human embryos because on this material there was the possibility to study the osseous labyrinth as a whole and its relation in space. We had the opportunity to do our investigations on the big collection of disarticulated embryonic skulls of the anatomy department of the medical faculty in Zagreb (Prof. Dr. D. Perović). This collection comprises 263 specimens.

The first results of our investigations were published in the monograph: "L'étiopathogénie de l'otospongiose et les facteurs anthropologiques" Archivo Italiano 1958, Supplement 68.

We hope that we succeeded in proving by these investigations that the human labyrinth repeats in its ontogenetic development the main stages of phylogenetic development.

We proved this by our observations concerning the shape and position of the oval and round window in space and the mutual position of the upper semicircular canals in the different stages of development.

We must stress that the evolution of the osseous labyrinth is not completed by the time of birth. The human labyrinthine capsule continues its transformation, reaching its definite shape by the end of the second decade.

Our investigations had at the beginning a pure scientific purpose, i.e. to enlighten the problem of ontogenetic development of the human labyrinth and it may eventually serve as a basis for the understanding of certain clinical problems. But analysing the forces that influence the shape of the labyrinth we came to the conclusion that there must exist an interdependency between the transformation of the osseous labyrinth and otosclerosis.

As we have been especially interested in this problem we continued our investigations concerning the evolution of the labyrinthine capsule in the fœtal and post-fœtal life.

New Observations

At the very beginning of our investigations our attention was drawn by the extraordinary configuration of the stato-acoustic nerve. We studied thereupon the anatomical relations in the opening of the internal auditory meatus. We came thereby to certain conclusions which verified our former observations and which shall be discussed here.

We begin with the description of the origin of the VIIIth cranial nerve from the brain.

The stato-acoustic nerve emerges from the brain at the inferior border of the pons lateral to the facial and intermediate nerves, as two roots. The medial and oral root belongs to its vestibular division, while the lateral and caudal one belongs to its cochlear division.

Leaving the brain the VIIIth cranial nerve shows a queer rotation in its course. This rotation is performed in such a way that the vestibular root passes between the pons and the cochlear root, crossing the latter. After this crossing the vestibular root is situated laterally and dorsally and the cochlear root, on the contrary, medially and orally. But this crossing of the two parts of the VIIIth cranial nerve is much more complicated than it seems at the beginning. While crossing the cochlear root the vestibular division shows a special rotation of its proper fibres on the right side in a clockwise direction and on the left side in the opposite direction. We can easily understand this rotation if we compare the position and shape of the vestibular division in the place when it emerges from the brain with its position and shape when it enters the internal auditory meatus. While leaving the brain it lies medially and is flattened dorso-ventrally. After the performed rotation it lies laterally and is flattened antero-posteriorly. Its medial fibres lie thus after the rotation cranially and its lateral fibres caudally (Fig. 3).

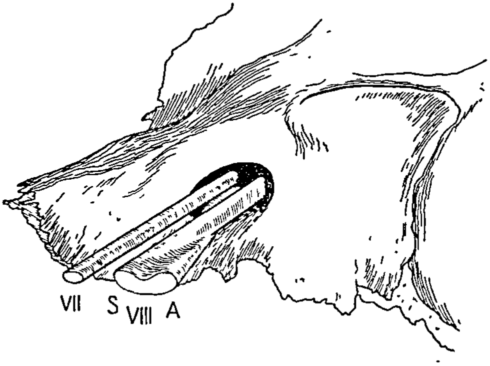

FIG 3

The double rotation of the two divisions of the stato-acoustic nerve Right side The originally medial, static (S) part of the VIIIth nerve lies after the performed rotation in the porus laterally and dorsally, while the cochlear (A) nerve, which was originally situated lateral to the former, lies after the rotation medially and ventrally Both divisions lie in the porus behind and below the facial nerve (VII)

The cochlear division must show likewise a rotation of its proper fibres in the same sense in order to keep its relation to the vestibular fibres. The cochlear fibres leaning against the vestibular nerve at the brain keep this position also at the opening of the internal auditory meatus. The lateral fibres of the vestibular nerve lie, as we have seen, after the mentioned rotation caudally. The medial fibres of the cochlear nerve which are related to them must follow their course; they rotate too so that, reaching the porus, they lie dorsally while the originally lateral fibres lie orally (Fig. 3).

The answer to the question why the fibres of the two divisions of the stato-acoustic nerve show such a complicated double rotation is to be sought in the shape of the fundus of the internal auditory meatus.

The fundus meatus acoustici interni is described in general as an oval field divided by a bony ridge, crista transversa, in an upper and a lower part. Crista transversa lies in an adult human fundus obliquely, being lowered at its posterior end. The bottom of the internal auditory meatus shows a system of holes allowing the nerve fibres to pass. In the upper part of the fundus there lies anteriorly the entrance to the Fallopian canal and behind it the superior vestibular area (area vestibularis utriculo-ampullaris) dotted with small foramina. In the lower part of the fundus there is situated anteriorly a curled tract of minute foramina (tractus spiralis foraminosus) that transmit the fibres of the cochlear nerve. Behind it there is a depression with minute foramina for the nerve twigs to the saccule (area vestibularis saccularis) and regularly a single foramen (foramen singulare) which transmits the nerve to the posterior ampulla of the semi-circular canal.

If we divide the fundus into four quadrants we can say that the two posterior quadrants belong to the vestibular nerve while the lower anterior quadrant lodges the cochlear nerve. The same relation of the fibres can be seen at the entrance to the internal auditory meatus. While entering the meatus the fibres of the vestibular division lie dorsally, i.e. by the posterior margin of the porus, while the cochlear fibres lie by its inferior margin. We can roughly delimit the two divisions of the VIIIth nerve by a vertical line drawn behind the facial nerve. The part of the VIIIth nerve lying behind this line belongs to the vestibularis and the part lying in front of it belongs to the cochlearis.

The double rotation of the two divisions of the stato-acoustic nerve is thus the result of its situation at the brain and of its position in the fundus.

The situation of the nerve fibres in the bottom of the internal auditory meatus can be explained by the phylogenetic and ontogenetic development of the labyrinthine block.

The position of the vestibular and the cochlear part of the labyrinthine block changes, as we have mentioned in the beginning, in the course of the phylogenetic as well as of the ontogenetic development, by rotation around the bi-temporal axis. The vestibular part is being shifted dorsally and downwards while the cochlear part is pushed ventrally and upwards. It is clear that the nerves are obliged to follow this rotation in the course of the phylogenetic as well as of the ontogenetic development.

We compared thereupon the fundi of the internal auditory meatus of the calf and dog with the fundus of adult man (Fig. 4). In the fundus of calf and dog we see also a bony ridge, crista transversa (Fig. 4, No. 5) dividing it in two fields and running obliquely in the antero-posterior direction. The anterior end of the crista is lowered and the posterior lifted. In the upper field there is the entrance to the facial canal (1) and the vestibular area (2). In the lower field there lies near the bony ridge foramen singulare (3) and a system of minute foramina for the cochlear nerve (4). If we keep the temporal bone in the right position we can see that the facial area is lying most anteriorly while the cochlear field lies below and behind it. The two divisions of the stato-acoustic nerve are situated, as we can judge by the position of the perforated area in the fundus, one above the other, i.e. the vestibular division above and the cochlear below.

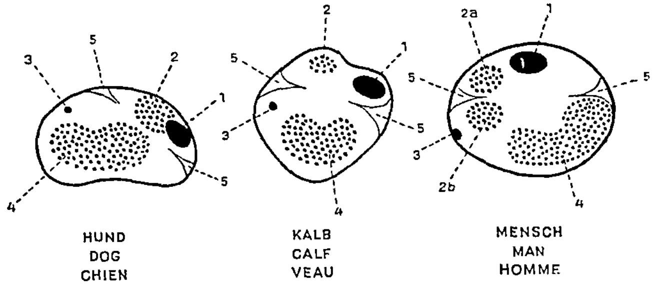

FIG. 4

The fundi of the left internal auditory meatus in calf, dog and man. One can follow the rotation of the minute foramina transmitting the parts of the stato-acoustic nerve in the course of the phylogenetic development The foramina transmitting the vestibular nerve (2, 2a, 2b) move backwards and caudally, while the cochlear region (4) moves ventrally and upwards. Crista transversa (5) changes also its position

If we compare these fundi with the fundus of adult man we can see that the transverse ridge (5) has changed its position. It is oblique but it runs in an opposite sense; its anterior end is lifted and the posterior end lowered. The cochlear area (4) moves forward so that it is situated in front of the afore-mentioned vertical line drawn behind the facial canal (1) and its anterior end lies 4 mm. in front of the entrance to the canal. The vestibular areas (1a and 2b) are moved caudally as well as the foramen singulare (3).

If we compare the fundus of the internal auditory meatus of the human embryos of 238, 262, 283 mm. and the one of the newborn with the fundus of adult man we shall see here all the stages of evolution repeated, although to a smaller extent (Fig. 5).

In human embryos the transverse ridge is not yet well developed so that the division of the fundus in two fields is still incomplete.

In the embryo of 238 mm. there can be seen the entrance to the facial canal (1). Behind it there lies a foramen for the twig to the utricle (2a) and in the region of the future crest there can be seen a hole to transmit the twig to the saccule (2b). A little more caudally lies the foramen singulare (3). The cochlear area (4) is represented in this stage by a single hole imitating the shape of the future tractus spiralis foraminosus. The cochlear area lies below the facial canal, its ventral end being situated 1mm. ventrally to the introitus canalis facialis.

In embryo of 262 mm. we see that the superior vestibular area (2a) is still represented by a single hole while the saccular area (2b) shows already a system of minute foramina lying in the region of the future crest. The cochlear area moves still forwards.

In embryo of 283 mm. the vestibular areas (2a and 2b) show on the left side minute foramina while on the right side there is still a single hole in the superior vestibular area (2a). The saccular area and the foramen singulare (3) are moved caudally. The cochlear area shifts with its anterior end still more forwards, so that it lies now 2 mm. in front of the facia canal (1).

In the newborn the transverse crest is well developed so that the two parts of the fundus are separated from each other. Above the crest there is the entrance to the facial canal (1) and the superior vestibular area (2a and below it lies the inferior vestibular area (2b), foramen singulare (3) and the cochlear area (4). The cochlear area moves still more forwards so tha only the part of it which corresponds to the basal part of the cochlea lie below the facial canal. The anterior end of the cochlea moved forward for about 2·5 mm.

We can thus follow the shifting of the labyrinthine block by change in the position of the transverse bony ridge, of the foramen singular and of the cochlear area.

The anterior end of the transverse ridge is lifted, as we have seen, in th course of development and its posterior end on the contrary lowered. Th change of the position of the transverse crest in the course of the phylo genetic as well as of the ontogenetic development is an indicator for th movements of the cochlear area and hence of the cochlea itself. Th anterior end of the bony crest is lifted by the anterior part of the cochlea region which is being shifted ventrally and upwards. This anterior end o

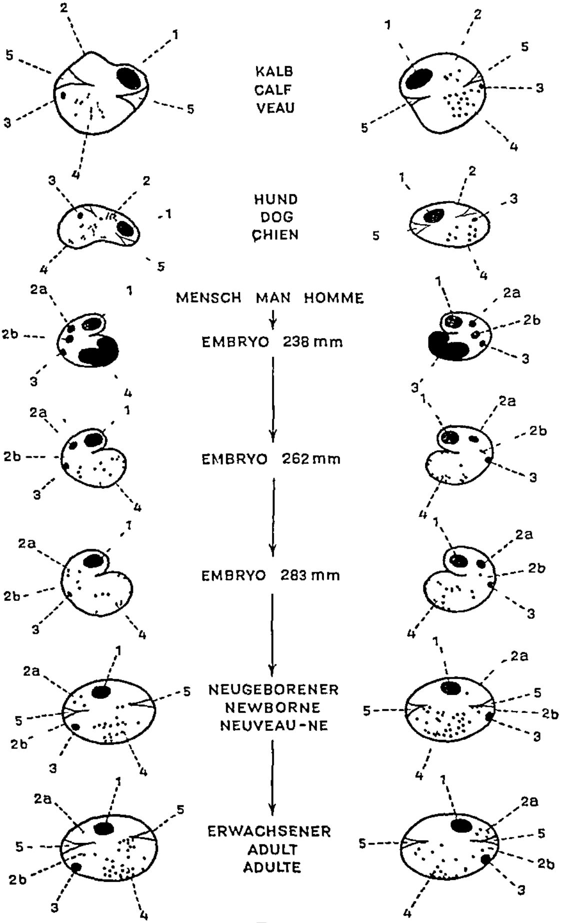

Fig 5

Represents the phylogenetic and the ontogenetic development of the fundus on the right and on the left side One can see the changes in position of the transverse crest (5) the shifting of the vestibular area (2, 2a 2b) and of the foramen singulare (3) backwards and downwards The cochlear area moves forwards and upwards so that it lies in adult 3 4 mm in front of the facial canal (1) the crest is lifted in the course of the ontogenetic development for about 15° (Fig. 6).

Foramen singulare moves caudally for 2 mm. In human embryos it lies about 0·5 mm. distant from the future crest, in the newborn it is about 1 mm. below it and in adult it is situated about 2 mm. below the crest.

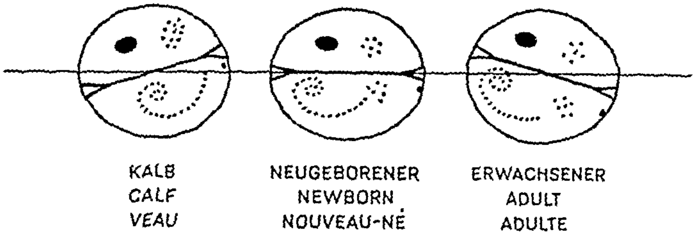

FIG. 6.

The changes of position of the transverse crest in the course of phylogenetic and ontogenetic development. The fundus of the internal auditory meatus. Right side. In the calf this crest runs obliquely in such a way that its anterior end is lowered and its posterior end lifted. In the newborn it has a horizontal course and in the adult it is lifted at its anterior end and lowered posteriorly. The transverse crest is thus the indicator for the movements of the cochlea forwards and upwards.

The moving of the cochlear area forwards can also be followed easily. The anterior end of the cochlear area moves in relation to the position of the facial canal ventrally for about 3 mm. In embryos of 238 mm. it lies about 1 mm. in front of the facial canal, in embryos of 283 mm. this distance becomes double i.e. 2 mm. In the newborn it is still bigger, 2-3 mm., and in the adult the anterior end of the cochlear area lies 3-4 mm. in front of the facial canal.

Comments

We could prove first that the special manner of rotation of the stato-acoustic nerve corresponds to the results of the phylogenetic evolution of the labyrinth. In vertebrates the labyrinthine block is situated so that the static part lies above and the acoustic part below the horizontal canal. In man the acoustic part lies anteriorly and the static part posteriorly. According to these topographic relations in man the acoustic part of the VIIIth nerve must be situated in the porus anteriorly or medially and the static part of it posteriorly or laterally. This crossing of the two parts of the nerve is the consequence of the changes in the topography of the corresponding end organs i.e. of the changes of the shape of the labyrinth in the phylogenetic development.

This statement is the starting point for the second question i.e. whether the bottom of the internal auditory meatus can give us proof of this. The topographic relations of the foramina which we find in the fundus as shown in Fig. 3 verify this statement. This can be seen most clearly in the fundus of the calf, where the minute foramina for the transmission of the twigs to the static organ lie above and for the acoustic organ below. In man the system of foramina for the static organ is situated laterally and for the acoustic organ medially The anatomical relations are, as we see, in harmony with the final stage of phylogenetic development

The third question which was for us of greatest interest is whether there exist the same ontogenetic changes in the fundus which we have found in the lateral wall of the labyrinthine capsule On this question too we can give a positive answer The fundi of the internal auditory meatus of human embryos of different stages represented in Fig 5 as well as the rotation of the transverse crest (Fig 6) show us that the foramina in the fundus belonging to the static part of the nerve move in the ontogenetic development backwards and downwards and the foramina transmitting the fibres of the acoustic nerve more forwards and upwards Even these minute observations in the fundus harmonize with the observations on the lateral wall of the labyrinthine capsule All that can be seen on the outer surface of the labyrinth, can be recognized also on its medial wall, the only difference is that in the fundus all verifying elements of the ontogenetic transformation are concentrated in a restricted space It makes the investigations more difficult but the results are, however not less convincing.

All these observations in the fundus complete and prove our former observations on the lateral wall of the osseous labyrinth We can thus conclude that our present observations prove our hypothesis that the labyrinthine capsule is repeating in its ontogenetic development the main stages of its phylogenesis We give herewith a positive answer to the question posed by Delattre

Conclusions

If we ask what are the forces responsible for the changes in the labyrinthine capsule in the ontogenetic development, we must come to the logical conclusion that these are the same forces that are responsible for the formation of the characteristic shape of the labyrinth in the phylogenetic development

The erect posture provokes not only the well-known changes of the spine but also changes of the base of the skull We mean herewith the angulation or the kyphosis of the base of the skull This process persists from foetal life up to puberty It provokes the action of the forces which influence not only the whole pyramid but also the labyrinthine capsule itself, changing its position and its shape We have found in the fundus of the internal auditory meatus facts which can prove that the labyrinthine capsule is transformed also in the course of the ontogenetic development We added to the doctrine of the ontogenetic development of the skull also the results of our own investigations concerning the individual development of the labyrinth.

But our present investigations have, according to our opinion, not only theoretic significance. In connection with the erect posture of man there persist as far as puberty certain mechanic forces inherent to the evolution of the skull. As the labyrinth is at this period already ossified there must appear in certain places which are more exposed to the influence of these forces, some changes in the structure of the bone, because the bone reacts to mechanical irritation in a specific way. If these changes occur in the places that are functionally important they can become manifest by clinical symptoms. This question will, however, be discussed in another place.

Acknowledgment

We are indebted to Prof. Dr. D. Perović, director of the Anatomy Department of the Medical Faculty in Zagreb for the free use of his precious collection of disarticulated skulls.

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FENESTRATION POST-MORTEM HISTOLOGY

By R THOMAS (York)

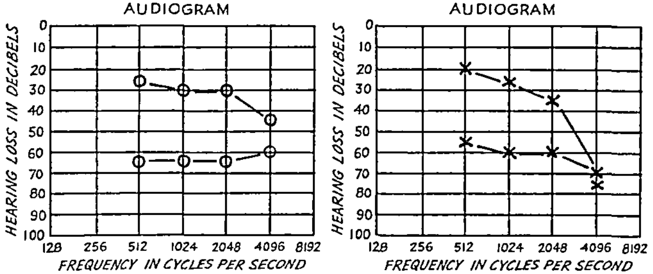
Two cases are presented Coronary thrombosis was the cause of death in one, and a road accident in the other

Case I Female, Schoolteacher, age 51

She died within a few hours of coronary thrombosis and the specimens were obtained following my chance perusal of the death notices in the local evening newspaper

Right ear In 1949 (she had been deaf for 30 years and wore an aid), a right fenestration was performed by the technique described by me in this *Journal* (1951) The operation was uneventful except that the meatal skin was found to be very thin, possibly as a result of this the fenestra was easily visible when this case was shown at a meeting of the North of England Otolaryngological Society in York in 1950 At that time the ear was already discharging and a small knob of cholesteatoma could be seen in the attic This ear continued to discharge until she died in 1956, but the fenestra was always visible The improved hearing level was retained

Left ear In 1951, despite the good hearing in the right ear, the patient asked for operation on the other ear This was done The mastoid was again very cellular, the stapes 'rock hard' and the meatal skin so thin that the fenestra became easily visible on this side too Despite every possible care, this ear started to discharge some months after operation and continued to do so despite every treatment



Audiograms showing best hearing improvement

The binaural hearing was so good that both she and her associates regarded it as normal. Furthermore, she stated that after the second operation she no longer had vertigo which, she said, had been due to turning her head so as to present her good ear towards her friends. She was delighted with her result and regarded her fairly frequent visits for aural toilet as a small price to pay for her practically normal hearing.

The histological pictures (coronal sections) should not only demonstrate the underlying condition for which the operations were performed and the mechanical reasons for their success; but also, since four years elapsed after the second operation, they should show the final reaction to the trauma of a successful fenestration operation.

Various Points Arise

1. *Thinness of flap*. This was noted at the operation and probably resulted in the fenestra being visible as a clear grey area. There is no calcification in the fenestra and the under surface of the flap is in places lined by endosteum and is sometimes adherent to the canal (Figs. 7 and 8).
2. *Edges of fenestra*. There is no greater regeneration of bone.
3. *Bone chips in the labyrinth*. There has been very little local reaction to some of these in the period of years, but in other places beyond the fenestra there is a considerable production of new bone and compression of the membranous labyrinth. It should be mentioned that this has taken place posterior to the fenestra and that it may have been due to the fact that I have hitherto not been too particular to remove all the chips from the posterior end of the fenestra. Bone dust can be seen embedded in fibrous tissue around the stapes (Fig. 8).
4. There appears to be little difference in the generalized otosclerotic foci which may be seen in both ears.
5. The development of *cholesteatoma* would appear to be related to the chronic infection within the remaining mastoid cells.

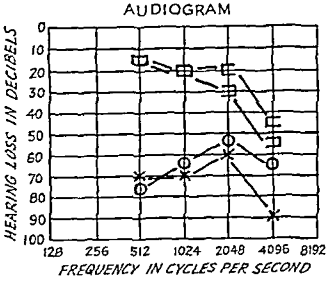
Case II. Male, Roadman, aged 49

He died in 1957 of multiple injuries shortly after a motor accident. I was lucky to hear of this and the specimens were obtained.

He had been deaf and his ears had both discharged continuously for many years until, in 1946, a suction clearance of bilateral aural polypi was performed and both ears became dry. There were, however, large perforations and each middle ear contained much scar tissue. The hearing (see audiogram) remained poor and the patient complained bitterly of bilateral tinnitus. Unfortunately, despite the good bone conduction, he proved intolerant of the Medresco aid which was also unsuitable for his work as a roadman.

In 1955 he pressed me to operate I performed an operation on the left ear which could now be described as a type 5 tympanoplasty The operation notes mention that both windows appeared full of scar tissue and it was thought that there was a *membrane covering* the round window (Fig 17). There was considerable attic erosion A fenestra was cut in the external semi-circular canal and a Thiersch graft applied, but not to the middle ear. An area of white bone was noted around the posterior part of the fenestra (Fig 14)

The result of this rather speculative procedure was not immediately good, the cavity healed well, but he had the most violent and prolonged post-operative vertigo that I have encountered He was certainly dizzy for three months The final result was, however, excellent, his hearing improved to a conversational level Unfortunately, as he lived 30 miles from York, his case was followed at a neighbouring Cottage Hospital where there is no audiometer There is therefore, no post-operative audiogram The statement that his hearing was good could be substantiated by the letter from his wife written, after he died, thanking me for his hearing improvement



In this case horizontal sections have been made of the cochlea and window regions The rest of the block was then re embedded and coronal sections cut to show the fenestra and skin-graft The unoperated right ear has also been sectioned The sections show that otosclerosis was largely responsible for the deafness, though infection has played a part The dislocation of the stapes in the unoperated ear (Fig 19) could have been due to rough treatment at suction clearance in 1946 In some of the sections (not shown here) there are small fractures and hæmorrhages due to the head injury In general the sections demonstrate the effects of combined otosclerosis and chronic otitis media If they are compared to the previous case it could be stated that the Thiersch graft has caused a little more reaction than the tympanomeatal flap The section (Fig 19) showing the cholesterol clefts in the stapes region and the dislocated footplate—in an ear that had been deaf and dry for nine years—is stimulating to one who is interested in both tympanoplasty and mobilization.

Summary and Conclusions

The histology of three persistent human fenestra novo-ovalis is presented. In two (from one patient) in whom both fenestra had been clearly visible, the fenestra had been covered with an exceptionally thin tympanomeatal flap. In the third the fenestra had been covered with a Thiersch graft. In the first patient the effects of otosclerosis and the subsequent infection of the fenestration cavities are demonstrated and in the other the results of chronic otitis media have evidently masked the unsuspected otosclerotic process.

To draw conclusions from two cases is premature, but further study of the sections (which are available) and comparison with subsequent material could be useful.

They do seem to support Lempert's (1951) contentions.

Acknowledgments

If these photographs have proved interesting, credit should be given, not to the writer who obtained the specimens by chance, but to the Ferens Institute of the Middlesex Hospital Medical School, London. I should like to thank Mr. J. P. Monkhouse, F.R.C.S., for his interest and help and Mr. D. Bishop, Senior Technician of the Institute, who spent so much time and trouble in preparation and annotation.

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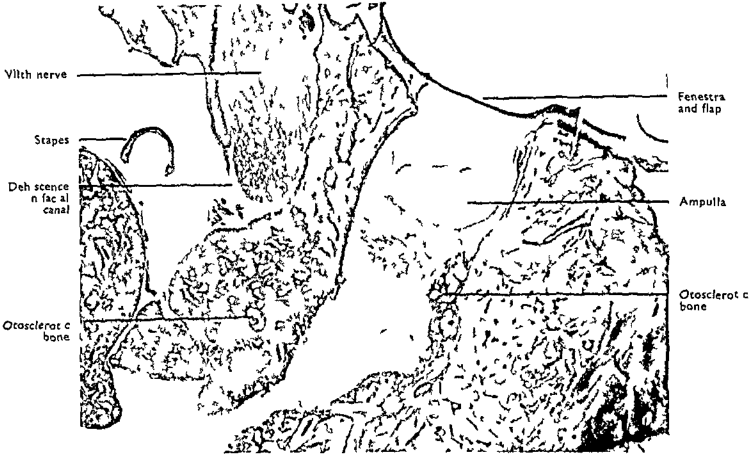

FIG 1

Case 1 Right ear Coronal section × 18 (approx) Showing fenestra anteriorly Note the coronal sections of each ear are in order antero posteriorly If the journal is held in the normal position the patient would be facing the reader The fenestra is therefore cut somewhat obliquely

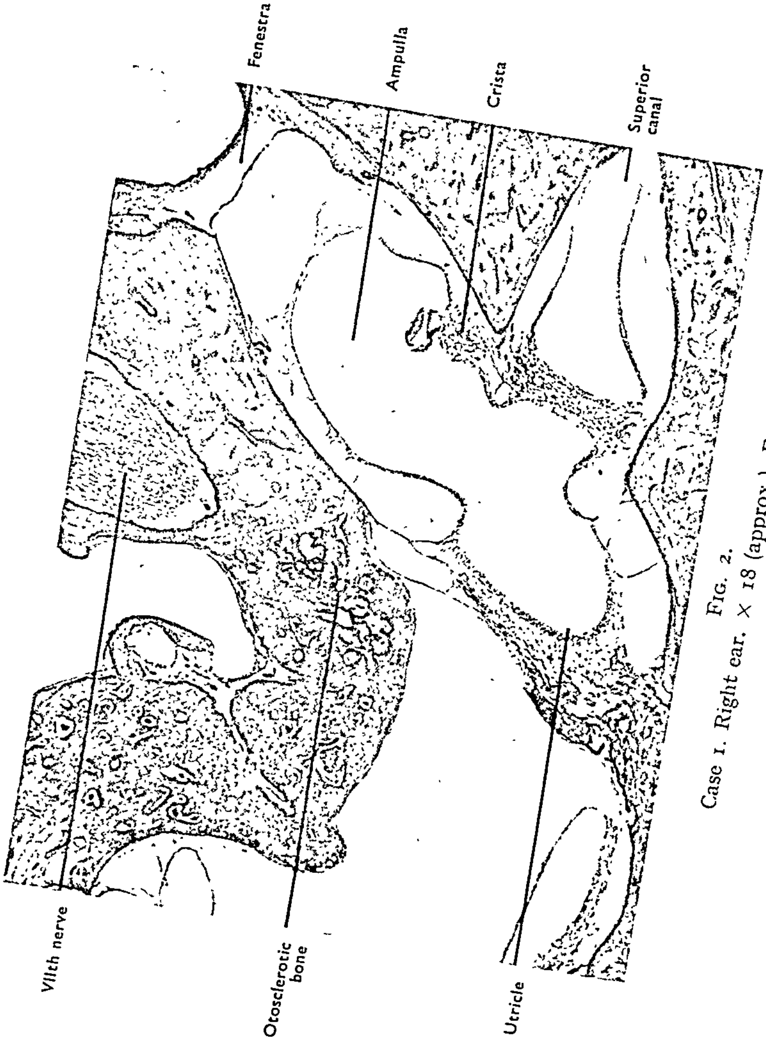

FIG. 2.
Case 1. Right ear. × 18 (approx.). Fenestra.

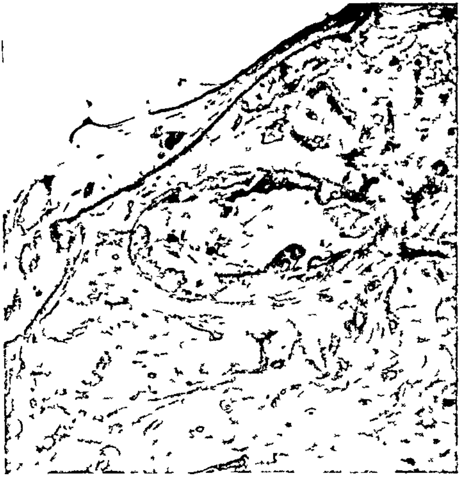

FIG 3

Case I Right ear Reaction in horizontal canal just posterior to fenestra New bone has formed and the membranous canal is compressed

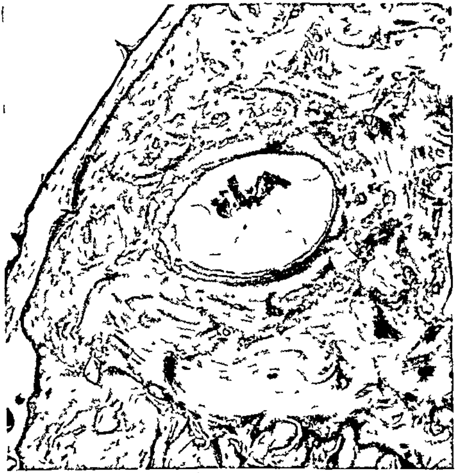

FIG 4

Case I Right ear Further back The canal is normal and there is very little reaction here to the bone chips after seven years

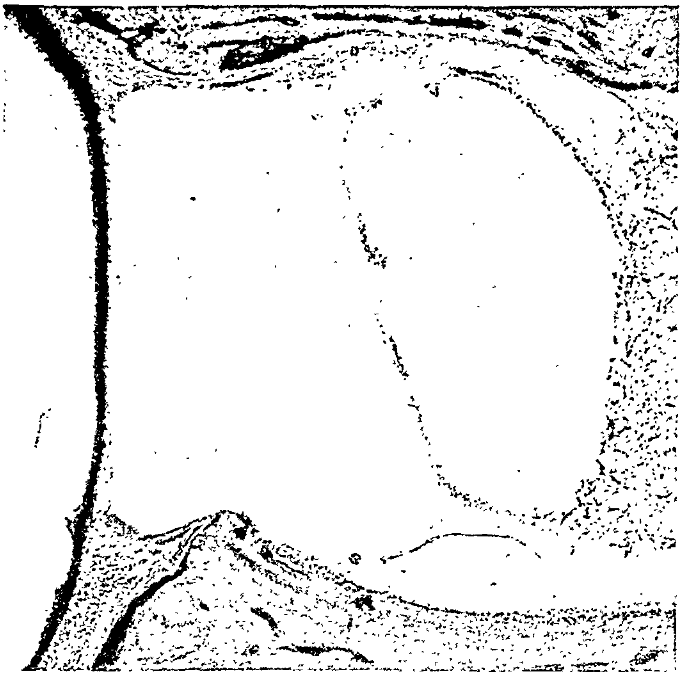

FIG. 5.
Case 1. Right ear. Fenestra × 44.

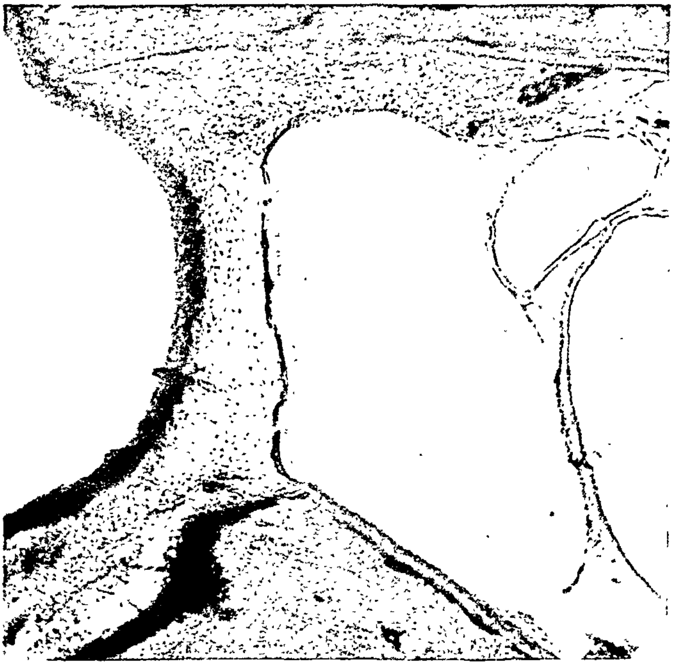

FIG. 6.
Case 1. Right ear. Fenestra × 44.

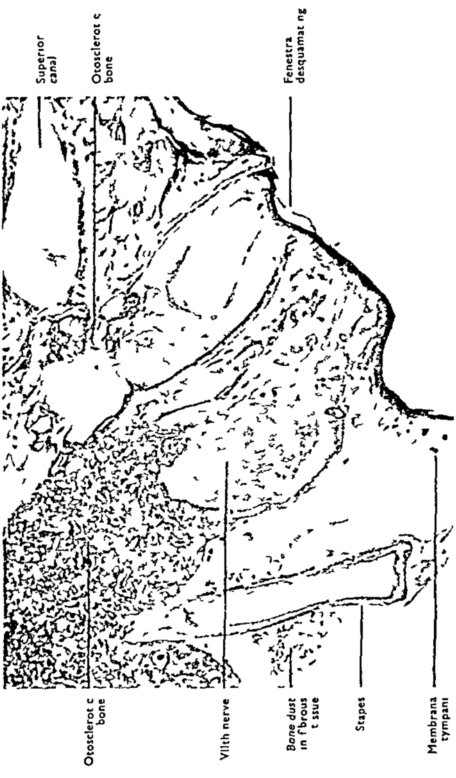

Fig 7

Case 1 Left ear Fenestra × 18 (approx) There is bone dust in the fibrous tissue around the stapes

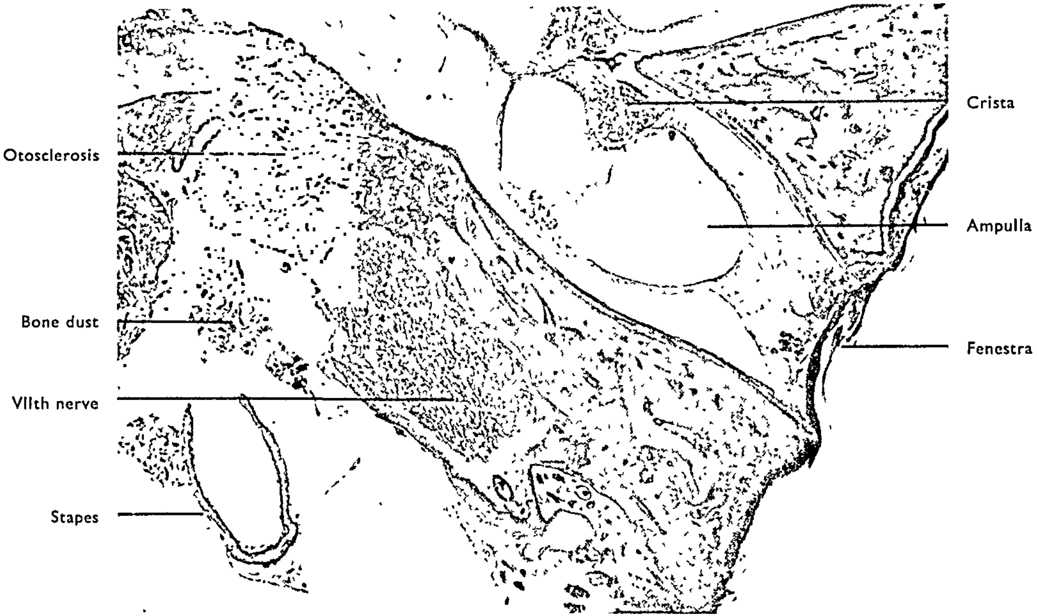

FIG. 8.
Case 1. Left ear. Fenestra × 18 (approx.).

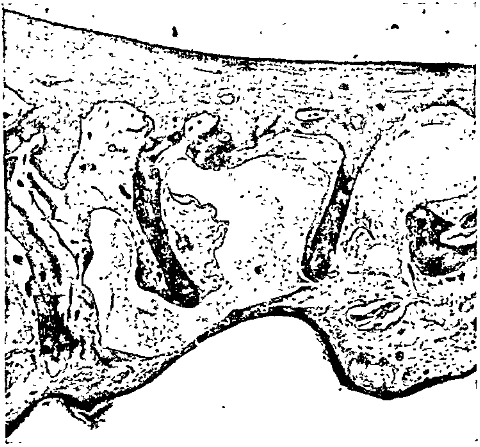

FIG 9.
Case 1. Left ear Cells in the roof of the cavity contain granulation tissue.

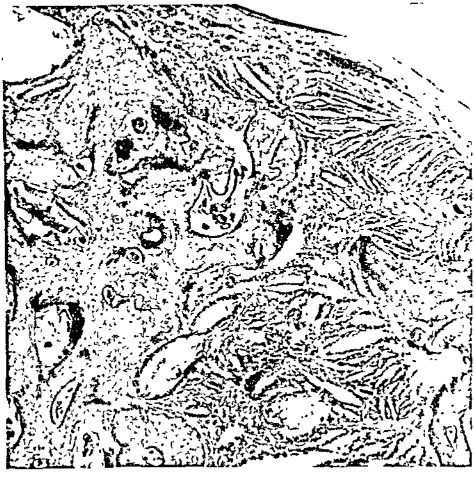

FIG. 10.
Case 1. Left ear. Cholesterol clefts and granulation tissue in fenestration cavity.

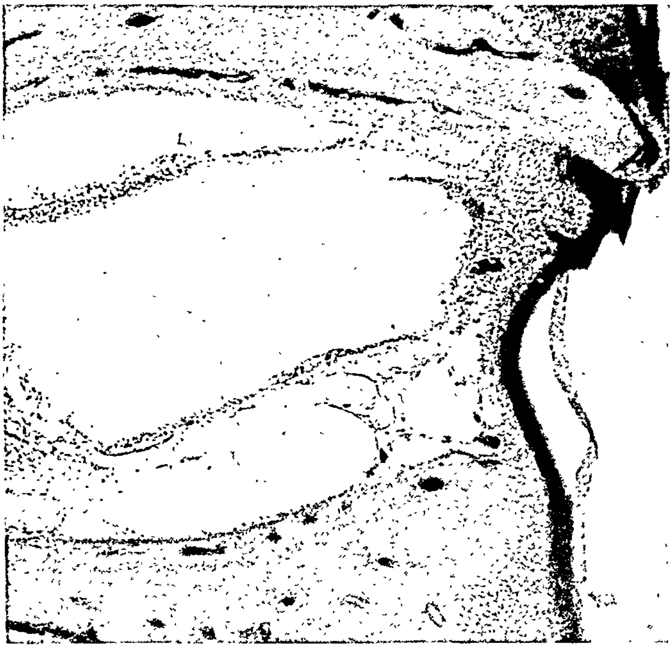

FIG. 11.
Case 1. Left ear. Fenestra × 44.

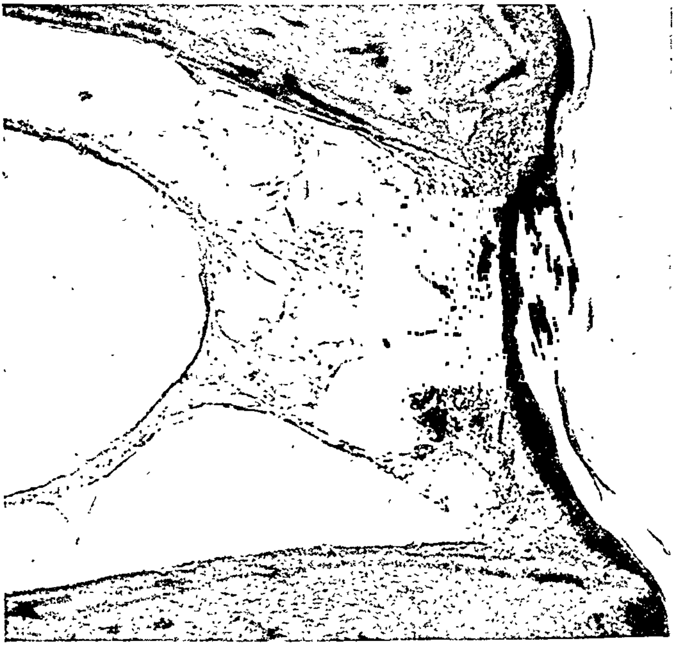

FIG. 12.
Case 1. Left ear. Fenestra × 44.

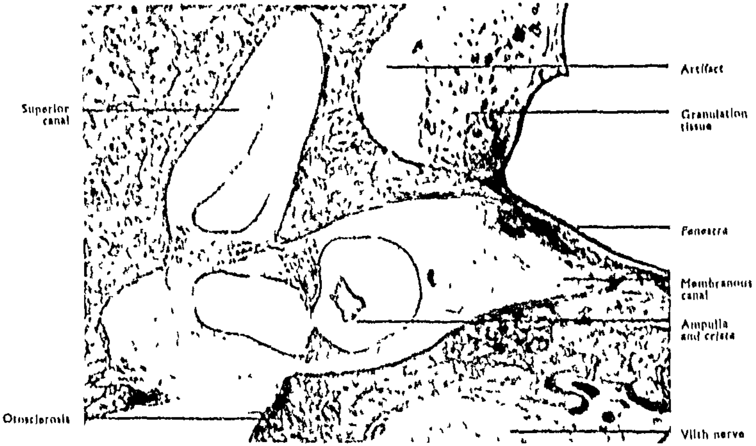

Fig. 13

Case 2 Left ear Fenestra and granulation tissue under Thiersch graft Normal orientation.

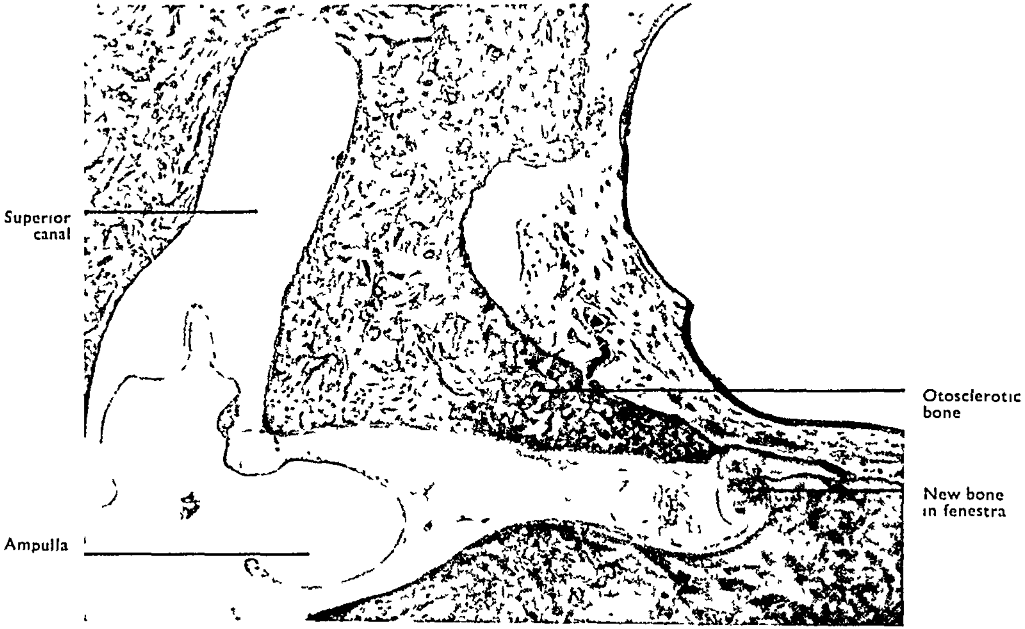

FIG 14

Case 2 Posterior part of fenestra closed by new bone with compression of membranous canal
Normal orientation

Fenestration Post-Mortem Histology

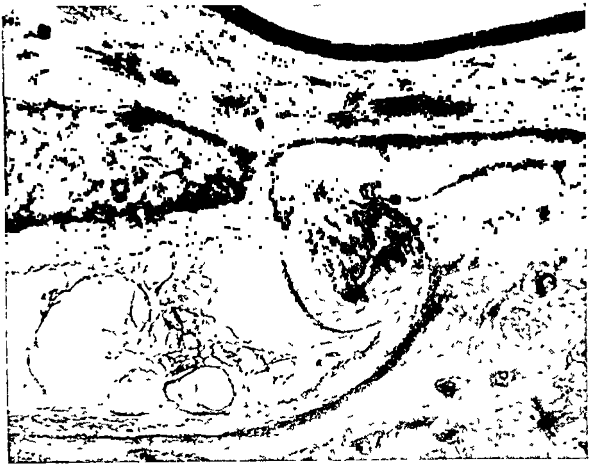

Fig 16
Case 2 Fenestra × 44 Showing closure

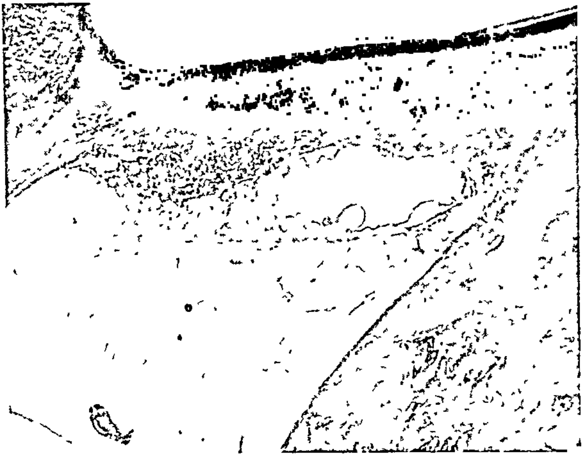

Fig 15.
Case 2 Fenestra × 44

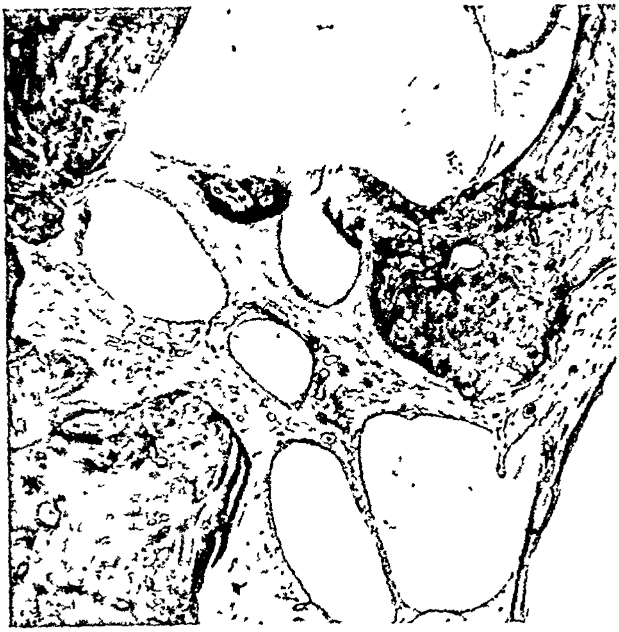

FIG 17

Case 2 Left ear Horizontal Section × 20 Cystic condition at round window

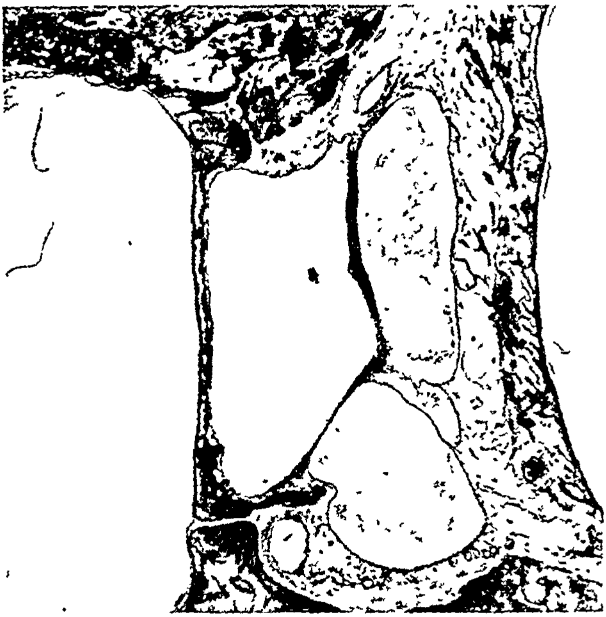

FIG 18

Case 2. Left ear Cystic condition underlying squamous epithelium at oval window.

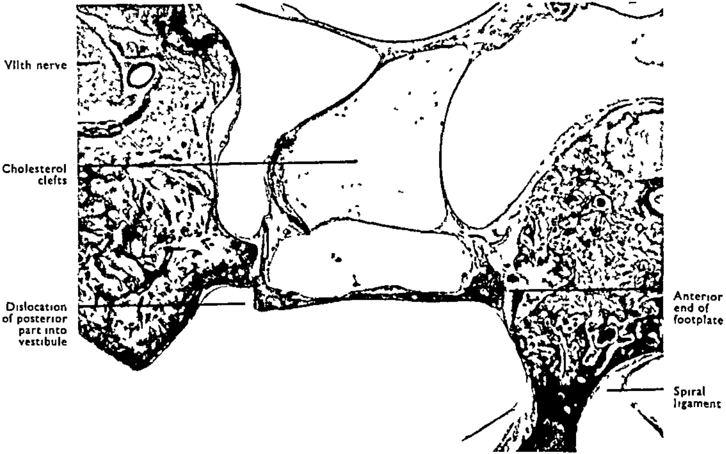
VIIth nerve
Cholesterol clefts
Dislocation of posterior part into vestibule
Anterior end of footplate
Spiral ligament

FIG 19
Case 2 Right (unoperated) ear Partial dislocation of stapes footplate posteriorly, otosclerotic fixation anteriorly and cholesterol clefts in cyst

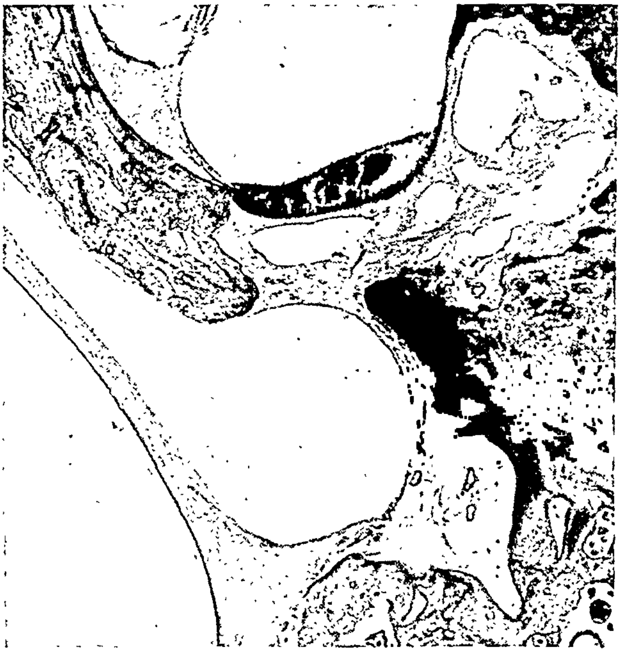

FIG. 20.

Case 2. Right ear. Lower part of drum and cystic condition of round window. Hæmorrhage due to head injury.

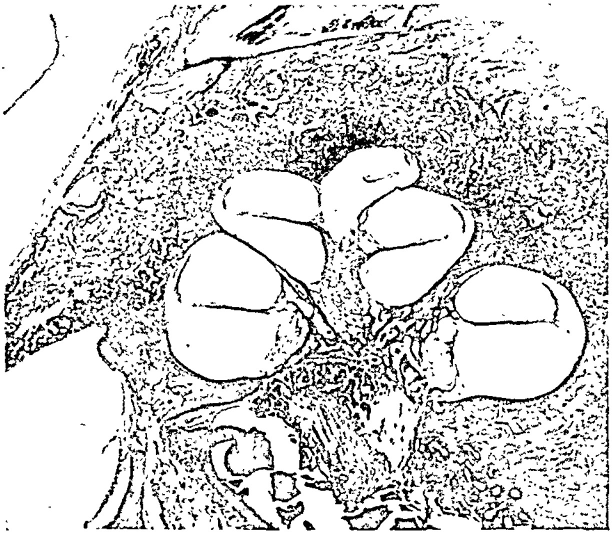

FIG. 21.

Case 2. Right ear. Cochlea × 10. Showing otosclerotic foci.

SOME OBSERVATIONS ON THE MOLE TECHNIQUE FOR DILATING ŒSOPHAGUS STRICTURES

By L. P. GRAY (Perth, Western Australia)

OVER the last 4 years, 6 cases of fibrous stricture of the œsophagus have been successfully treated by the mole technique at the Princess Margaret Hospital for Children, Perth, Western Australia. The method is as reported by Harvey and Negus (1953), but some modifications in the technique and apparatus used have been evolved (Gray, 1957). In this technique, hollow dilators or moles, of varying size are drawn by strong thread, per os, through the œsophagus and out at a gastrostomy. The thread is tied to elastic which is attached to the lower abdomen. This exerts continuous gentle traction on the mole, which dilates the stricture as it passes. No anæsthetic is required for the insertion of the mole, moderate sedation being sufficient. This paper reports some further observations on the technique.

The treatment of strictures of the œsophagus is usually a combination of any of the following:

(1) Dietary: fluids or soft mushy foods.
(2) Indwelling flexible hollow tubes:
  (*a*) Nasal tube into stomach.
  (*b*) Souttar tube.
(3) Dilatation:
  (*a*) per os—
    (i) Blind bouginage.
    (ii) With swallowed thread as a guide.
    (iii) *Viâ* œsophagoscope.
  (*b*) Utilizing gastrostomy:
    (i) Using retrograde bobbins or Tucker bougies.
    (ii) The mole technique.
(4) Surgical:
  (*a*) Local excision.
  (*b*) Anastomosis and short circuit operations.

The permanent alleviation of symptoms is usually best achieved by early efficient dilatation. For dilatation to be efficient it must:

(1) Be gentle—to prevent local tissue reaction.
(2) Act for as long as possible—to keep the fibrous tissue stretched.

(3) Cause maximal dilatation of the stricture—to allow for the inevitable fibrous tissue contraction later.
(4) Be safe—the risk of perforation to be minimal.
(5) Cause minimal psychological trauma (particularly applicable to children).

The mole technique fulfils all these criteria. It also has an advantage over retrograde bouginage in that the discomfort of pulling dilators and taut thread over the sensitive epiglottis and back of the tongue does not occur.

The indications for use of the mole technique appear to be:

(1) Strictures requiring very gentle dilatation—i.e. those in which rupture or perforation are likely to occur.
(2) Strictures requiring repeated and frequent dilatation over a prolonged period (say for 1 to 2 years); including those in which repeated anæsthetics are contra-indicated and those in which psychological trauma must be kept to a minimum.
(3) Failed adequate dilatation by other methods.

The moles, or dilators used, vary from those described by Harvey and Negus in that they are cone-shaped instead of pear-shaped, the lumen are larger, are made of smooth polished acrylic and contain a metal marker for X-ray purposes. The smooth cone shape permits gentle progressive dilatation. The sizes used range from 20-50 mm. in length and 10·5 to 18 mm. diameter—larger sizes have not been used because only small children have been involved.

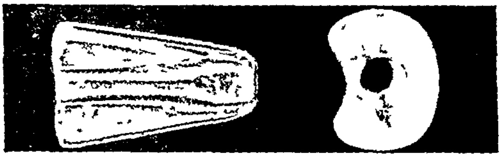

FIG 1

Indented moles (natural size) For use in post-cricoid strictures

Progressively larger moles are inserted up to the maximum size corresponding to the age, although a child of 2 years has been dilated to take an 18 mm. diameter mole without any apparent ill effect (the normal post-cricoid diameter for this age being 12-14 mm. (Gray, 1957)). Whenever possible the size is selected so that the mole does not take more than 5-6 days to pass. When the time of passage is down to 2-3 days, the size of the mole may be increased. The time between insertions should be at least about 10-14 days. This ensures that any reactionary œdema that may have occurred will have subsided. The time interval is lengthened towards the end of treatment. Œsophagoscopy has been performed on several occasions immediately after the passage of a mole, but no excoriation, necrosis or break in the mucosa has been found except to a minor degree on one occasion

Some strictures are very elastic, and though they dilate fairly readily, they rapidly recur These require the frequent, slow passage of large dilators to produce over-dilatation The time of passage can be lengthened considerably by diminishing the tension exerted by the elastic traction The size of the œsophagus can be assessed by passing rubber bougies (such as Hurst mercury bougies) The gastrostomy should not be closed until a 13 mm bougie can be readily passed

Fig 2

A case of congenital tracheo œsophageal fistula (common type)—lipiodol swallow æt 2 days

The lumen of the moles which vary from 3 5 mm to 5 mm in diameter allow a considerable range of diet to be taken orally, such as breakfast cereals, biscuits and most puddings The main meal, which must be reduced to a fine homogenous pulp as in a 'vitamizer , is not very palatable, and is usually given *viâ* a catheter through the hollow gastrostomy plug without altering the tension

The high post-cricoid stricture presents many problems. In particular, there is no reservoir effect above to assist dilatation, and the A.P. diameter is so reduced that even a moderate sized dilator may cause tilting of the cricoid, with laryngeal embarrassment, thus preventing the use of larger sized dilators. A series of flattened, indented moles are now being used (see Fig. 1) which have been a great help with these cases. With A.P. diameters of only 10-12 mm. and transverse diameters of 15-18 mm. these correspond in size to 14·5-17 mm circular moles.

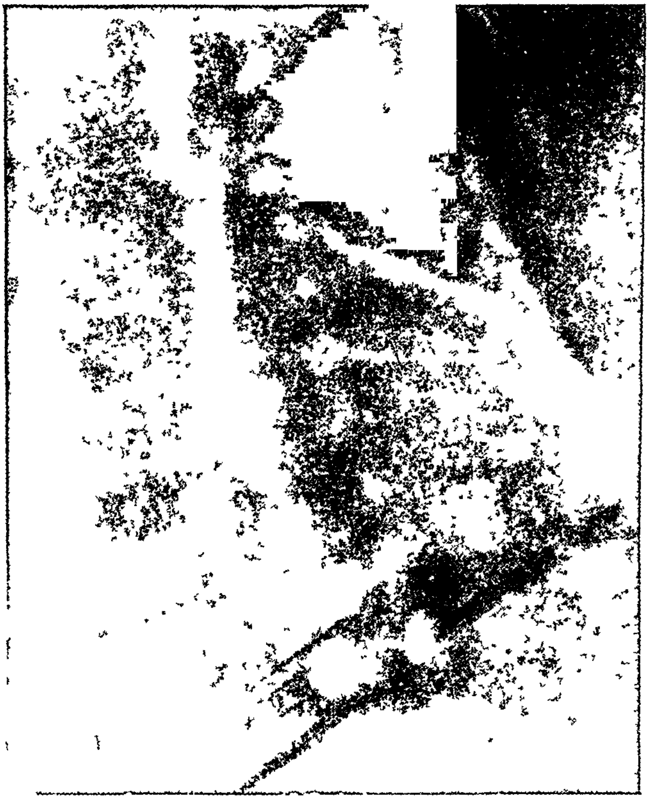

FIG 3
Showing complication of fistula formation 12 days after operation

The gastrostomy plugs as described by Gray (1957) have been found to be very efficient, and prevent excoriation of the abdomen. The hollow gastrostomy plugs help to protect the cardia and allow a more direct pull down the œsophagus The solid plugs have also been used with satisfactory results in adults with gastrostomies It is considered now that the plugs should be about 16 mm in diameter, for this keeps each gastrostomy opening large enough to allow easy removal of the mole from the stomach.

The gastrostomy has been closed in 3 cases due to caustic soda burns, and in one due to reflux œsophagitis following hiatus hernia The other 2 cases—a double stricture due to caustic soda burns and a case due to complications following surgical repair of a congenital tracheo-œsophageal fistula—are nearing the time for closure of the gastrostomy

The treatment after closure of the gastrostomy has been the passage

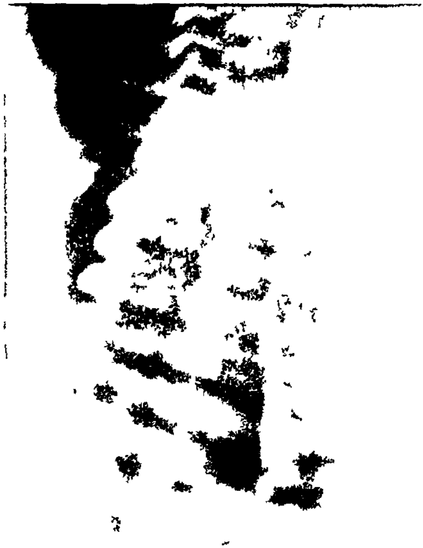

Fig 4

Aet 3 months showing tortuous narrow stricture before commencement of dilatation

of rubber bougies (up to 39 or 42 French grade) every 6 to 12 weeks (using a ¼ or ½ of an amethocaine lozenge as local anæsthetic) The passage of rubber bougies causes little actual dilatation of a stricture and ensures the early recognition of any diminution in the size of the lumen No further dilatation has been necessary in the burns cases and 2 of these have been closed for 2 years The hiatus hernia case has had no reflux since operative repair of the hernia, but gets intermittent spasm which is relieved by the passage of rubber bougies

The following case report demonstrates the adaptability of the technique:

An infant had a congenital tracheo-œsophageal fistula repaired when 3 days old. The deformity was the common type of blind upper pouch, with the distal end of the œsophagus opening into the lower trachea (Fig, 2). The distance between the two ends of the œsophagus was such that the surgeon (Mr. D. W. Fleming) was only able to make the end to end anastomosis with a single layer of interrupted sutures. The chest was drained and a gastrostomy performed. A fistula occurred which drained along the path of the drainage tube for about 3 weeks (Fig. 3). A stricture then developed at the anastomosis site, but he was able to be breast fed.

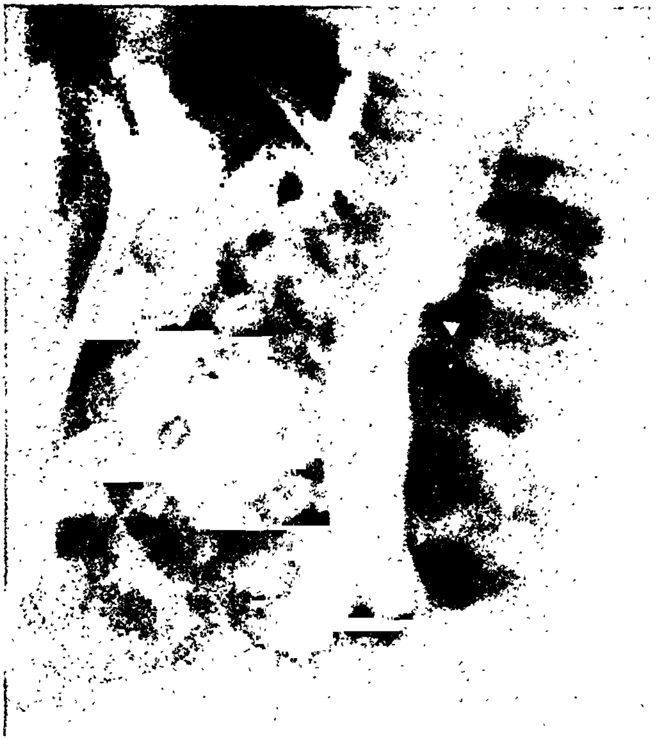

Fig. 5.

Aet. 10 months. Practically no sign of stricture—arrow at site of previous stricture.

When aged 3 months, a lipiodol swallow (Fig. 4) demonstrated considerable stenosis. Œsophagoscopy was performed and dilation of the narrow tortuous stricture was begun by using "ferret and follower" urethal dilators. The mole technique was then commenced using small moles, and he progressed satisfactorily until aged 6 months when he began choking over cereals. Œsophagoscopy revealed considerable narrowing and a thin diaphragm type of stricture with a small central hole. The size of the moles was rapidly increased to a greater size than the normal œsophageal diameter for his age, so that at the age of 10 months an indented mole equivalent to a 15.7 mm. diameter dilator was passing through easily in 48 hours, and the X-ray (Fig. 5) was very satisfactory. The indented moles were used because these pass more readily through the post-cricoid area.

When aged 11 months, he was well, but found to have a hæmoglobin of 7.5 gm. and was given iron therapy and 350 cubic centimetres of blood. His blood picture has been normal since. Recent œsophagoscopy (when aged 15 months) revealed only a short white band in the posterior segment of the œsophagus as evidence of the previous constriction. A 16 mm. diameter mole passed easily in 48 hours. His weight then was 19 lb. 10 oz.

It is considered that he will require very little further dilatation.

Conclusion

This technique is very safe, efficient, causes minimal psychological trauma and is particularly suitable for small children.

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REPAIR OF TYMPANIC PERFORATIONS

By J. F. O. MITCHELL (Dundee)

So much attention has been focused recently upon operative repair of the tympanic membrane that some of the older well-tried methods are apt to be forgotten; brought up to date they still hold an important place in our choice of treatment.

The purpose of the present communication is to show how much can be attained by conservative means.

Persistent tympanic perforation has for long been a challenge to medical science. It must result in some loss of hearing, for the drum area is decreased and protection of the round window diminished, so that the differential required to produce perilymph movement is reduced (Ormerod and McLay, 1956). Again some degree of fibrosis and adhesion is a not uncommon concomitant and will limit drum, ossicular and window-membrane movement: this latter is not remediable by repair of the drum membrane.

In addition, however, there is the constant liability to infection of the exposed middle-ear mucosa, with recurrent otitis media, at least a nuisance, at most a danger to life from complications.

The social aspect, however, is equally important—a child whose parents must exercise care with every hair-wash or cold, who cannot go swimming with the others, or must attend surgery or clinic for frequent attention to otorrhœa, is very "ear-conscious," feels different from other children, and the psychological aspect is as important as the acoustic.

As a young adult again he may find himself unfit for Military Service; or find that otorrhœa interferes with efficient use of a hearing aid.

Again eustachian catarrh may be the sole source of persistent otorrhœa after an otherwise satisfactory modified radical mastoid operation.

Attempts to deal with this situation have been generally along one of three lines:

(1) Protection of the exposed middle ear, varying from simple care and use of a wool plug, to Banzer's ivory tube covered with a membrane of pig's bladder (1640), or Schaffer's Acrylic seal to allow even swimming (1956).

(2) Relief of the associated deafness—from a simple pledget of cotton wool (Yearsley, 1857, Asherson, 1950) which in a sub-total defect restores the differential window impedance, or a rubber disc

(Toynbee, 1857), or sized paper disc (Blake, 1887), or cellophane disc (Nasiell, 1934), applied over the perforation, to an acoustic probe of nylon inserted through a fish-skin diaphragm stretched over a plastic cylinder moulded to the meatus (Pohlman, 1947) At best these methods are serviceable only with access to an otologist, and at worst lead to recurrent otitis media or externa

(3) Repair of the drum defect—from
   (*a*) Stimulation by repeated cautery, or
   (*b*) Stimulation by cautery followed by the application of an inert prosthesis to act as a scaffolding for the ingrowth of epithelium, to
   (*c*) Skin graft repair

This last line of approach, repair, would seem to be the most logical, and certainly has appealed to most workers There is no doubt that a defect of the drum membrane means no resistance to the passage of air, mucus and therefore infection, up the eustachian tube from the naso pharynx on nose-blowing, thus effectively preventing its own repair

Failure of a perforation to heal spontaneously is accepted as due to epithelialization of its margin by the outer squamous layer's growing inwards to meet the mucous layer, likening the perforation to a track or fistula This epithelialization is usually the result of chronic otitis media although the large defect produced by acute necrotic otitis or by blast rupture may be a factor, the epithelial 'track' must be completely destroyed before connective tissue can re establish continuity

Acid cautery, as well as destroying the epithelium, incites an inflammatory response in the middle layer of connective tissues with fibroblastic proliferation laying down collagen fibres which, in shortening, may facilitate closure by purse-string action (Baron, 1948)

There is divergence of opinion on which layer plays the dominant role in closure Stinson (1936) suggests the outer squamous epithelial layer, and although this may explain those which heal by thin transparent atrophic scars, the full thickness invisible repair produced by acid cautery of the margins suggests that all three layers play an active part, although some scarring may result from the two layers of elastic tissue being replaced by less specialized fibrous tissue with greater powers of regeneration certainly the inner mucous layer carrying the increased blood supply to the edge of the perforation is of extreme importance (Hall, 1951)

The vitality of the epithelial layer is well seen in the migratory or escalatory (Schrimpf, 1954) effect as proved by the migration of patches (Stinson 1941) or Blake s discs from the site of perforation usually to the posterior wall and shown by the change in size, shape and position of perforations during healing (Korkis, 1946) this was first recorded by Stinson (1936) in 1932 who noticed the progressive shift of a foreign body embedded in the epidermis, and confirmed it subsequently by ink markers; this migration accounts for the normal elimination of detritus from the deep meatus (Simonetta, 1947) and its failure for keratosis obturans: again failure to heal can be attributed to doubling back of the epidermis on the substantia propria.

Recently Diamant (1954) has suggested that the possibility of spontaneous cure can be told in advance, by correlation with the cellularity of the mastoid, a larger cell system giving more chance of spontaneous cure.

Roosa (1876) and Politzer (1894) used silver nitrate to cauterize the epithelial edge, and Okuneff (1895) was the first to use trichloracetic acid which remains the favourite method today. By repeated application, Adams (1952) has closed 10 out of 12 perforated drums with audiometric improvement. Other workers have similar results at the expense of unlimited patience (Dunlap and Schuknecht, 1947). Other methods include scarlet-red ointment (Unger, 1947), and hot air applied *viâ* a eustachian catheter (Griot, 1931), but all these had the disadvantages of a large number of treatments and frequent otitis.

Cautery followed by "scaffolding", inert or otherwise, has been the most successful method. Joynt (1919) was the first to use this combined method in four successful closures with trichloracetic acid; Linn (1944) followed the acid with "protonuclein dusting powder" on cotton and "euthymol" drops, while Stinson (1941) and Fox (1945) followed with Cargill's membrane (sheep's mesentery). More recently Schrimpf (1954) has used prepared amniotic membrane which he found adhesive and transparent, closing 48 of 53 central perforations. Although the transparency was an advantage in watching progress, the preparation of the membrane is admittedly "tedious and involved".

Surgical repair was proposed by Berthold (1879) who applied adhesive plaster to the perforated drum for 3 days, then applied a skin graft from the arm; and a modified technique was revived by Shulhof and Valdez (1944) quoted by Derlacki. Reading (1946) suggested the use of thrombin as an adhesive for split-skin grafts, while Frenckner (1955) and Hall (1955) suggested the use of a full thickness pedicle flap; Zöllner (1955a) however, found these more difficult and just as liable as free grafts to necrosis and perforation. He abandoned them in favour of full-thickness post-auricular grafts after scarification of the margins or meatus, under antibiotic cover (Zöllner, 1954). Further experience in this work with collar incision of the perforation is recorded by Ormerod and McLay (1956). House (1953, however, prefers radial incision of the drum remnant prior to grafting.

Adhesion of the graft to the promontory is essential for its nourishment and survival in large defects, and this is encouraged by scarification or turning down a mucosal flap; but a continuous cavity from the eustachian tube to the round window is essential for good hearing and this is encouraged by packing with silk thread (Zollner, 1955b) or with gelatin sponge (Gisselsson, 1954)

Although freedom from recurrent otorrhœa and improvement in hearing are the chief aims in treatment, it is useful to obtain beforehand some idea of expected results I have seen bilateral sub-total defects of some 40 years' duration in a street orderly who had no knowledge of otorrhœa, and in such cases a patch prosthesis, or the acoustic probe, mechanical (Pohlman, 1948) or audiometric (Bennett, 1956, Ormerod and McLay, 1956) will tell whether any hearing improvement will justify interference Again a good audiogram by bone conduction healthy windows, and a patent eustachian tube are essentials to success (Pietrantoni, 1954) More than 40 decibels loss by air conduction up to 2048 c p s suggests disease of ossicles or temporal bone (Fox, 1945) It would seem possible therefore, that closure of any tympanic perforation (excluding marginal ones with epithelial invasion of the middle ear, or those with active disease, in both of which cases the more extensive attico-antrotomy with tympanoplasty is indicated) is today possible often with the minimum of time and trouble for surgeon or patient

I have used the trichloracetic acid cautery method but combined with autogenous blood clot as suggested by Biber (1954), I found however that transference of the clot to the drum was very difficult, and having already tried dry gelatin sponge as a patch prosthesis, I combined the two and soaking the piece of sponge in autogenous blood, found a moist pliable patch, easy to manipulate, and giving ready estimate of hearing improvement This would appear to simulate more closely nature's method of organizing the blood clot or exudate which fills a recent perforation

Technique

Success has been achieved only with perforations which have been clean and dry for several months and in which there is no associated middle-ear or eustachian catarrh The outer meatus is cleared of any wax and debris and painted with surgical Dettol (Reckitt and Colman) The entire margin of the perforation is carefully touched with a fine tightly wound wool applicator dipped in 50 per cent trichloracetic acid and any excess removed (50 per cent was the concentration found by trial and error to give best results with least pain) taking care that no acid touches the medial wall of the middle ear No local anaesthetic was found necessary in the great majority of cases, even older children, as Adams (1952) agrees, although Schrimpf (1954) used Greig's Solution (10 per cent. cocaine in equal parts of 95 per cent ethyl alcohol and aniline oil) to sterilize and anæsthetize the drum and produce a desired hyperæmia In others 20 per cent *cocaine in aniline oil proved adequate*

With full aseptic technique a small square of Sterispon gelatin sponge

(Allen & Hanbury) is soaked in the patient's own blood. Originally this was done by thumb-prick after scrub and surgical Dettol, the gelatin sponge being laid over the puncture wound until sodden, then lifted into a sterile aural speculum; latterly, however, a thumb-prick on occasions did not give adequate blood, and venepuncture of 1 c.c. by syringe into a sterile receiver, was used instead.

The sodden square is under direct vision lifted by forceps from the speculum and placed over the perforation, then manipulated into position using a Jobson-Horne probe, pressing it down firmly over the whitened edges.

A small wool plug is inserted in the outer meatus and the patient instructed not to blow the nose, for 48 hours.

In the majority of cases no further treatment is necessary, the patch clotting firmly adherent to the margins of the perforation and remaining so as the defect heals and the patch migrates to the posterior wall. There is never any need to remove it from the drum; but when the whole drum is seen intact proximal to the now hard black patch on the posterior meatal wall, it can be gently lifted off, although in many cases it will fall free of its own accord.

If the first attempt should not succeed, yet the perforation and middle ear remain clean and dry, the margins should be cauterized again on two or three occasions, and then the patch repeated; if the middle ear is moist the perforation may not have been ready for closure and attention should be directed towards clearing any infection there, or in the nose and throat, after which a period of observation, and further treatment can be commenced.

Large central or sub-total defects may not respond to this conservative method. Like many others, I have tried the application of split skin grafts with little success even under antibiotic cover. The work of Zöllner and Pietrantoni on full-thickness grafts suggested a fresh attack. The marginal drum remains may be scarified or incised but should not be sacrificed as the original tissues are much more resistant (Zöllner, 1955b); the collar incision of Ormerod and McLay (1956) has proved effective, but in total loss scarification of the meatus or circular incision (3-4 mm. proximal to the annulus) and turning inwards a full-thickness collar will supply a raw area for nutrition of a free-full-thickness graft. Beales (1957) has since described a similar technique. Additional nourishment can be obtained by turning down mucosal flaps from the promontory over the gelatin sponge soaked in appropriate antibiotics, usually crystamycin, to maintain a "window-tunnel". The post-auricular full thickness graft is accurately cut (with the help of a "template" of oiled silk), thinned peripherally, fitted, and packed in position with further gelatin sponge crystamycin, kept moist by a seal of oiled silk, while systemic cover is maintained by crystamycin injections for 8 days, the patient being warned against blowing the nose. Again there is no need to remove the gelatin sponge packing, as, with removal of the oiled silk it dries up and crumbles away. Thereafter every care should be exercised to avoid ascending infection from the nasopharynx with sloughing of the graft. Access in some cases is limited by stenosis of the meatus, but this can be overcome by endaural incision.

While this surgical method requires admission to hospital for 9 days or so, I do not feel it is any more certain of success in moderate defects than the acid cautery and autogenous sponge technique (although Beales finds acid cautery alone tedious and likely to produce a scarred and rigid drumhead); and as this requires only out-patient attendance and no anæsthetic, it is much preferred by the patients, even if repeated attendances should be required.

Results

Cases with active suppuration, acute or chronic, or with posterior marginal defects, were considered unlikely to benefit from the treatment.

Treatment has been undertaken in 50 cases which were thought suitable, and of these 36 have healed rapidly by the conservative cautery and sponge-clot technique, and have remained intact since. Of these 36 cases, 14 drums healed after a single treatment, 6 after the second, while 16 required repeated cautery (average 4·25 times) before success was achieved.

Six further patients defaulted after one or more treatments, and have not since reported for examination.

During the period of this investigation, 5 cases were seen in which persistent perforation of the drum healed spontaneously following relief of some preventive factor.

1. Cautery of granulations on the promontory followed by Aurist. Acid Boric (1 case).
2. Stimulation of granular mucosa by Aurist. Acid Boric (1 case).
3. Control of local infection, by Aurist. Chloramphenicol (3 cases in one of whom adenoids were removed also).

This serves only to prove that, given the opportunity, nature will herself remedy the defect.

Eight drums failed to heal under treatment and of these 5 had no complaint of any sort and as they did not respond to one or two treatments, further persistence was hardly justified; in some of these, adhesions may wall-off the middle ear without impairing hearing.

Two will require surgical treatment (tympanoplasty) for active chronic suppuration, which was not at first apparent.

One will require surgery for access impaired by meatal exostoses.

Healed	36	14 after 1 treatment. 6 after 2 treatments. 16 after repeated treatments.
Defaulted	6	
Persisted	8	5 abandoned. 3 require surgery.
	50	

Drums which required repeated treatment were those with large or with anterior (possibly marginal) defects difficult of access.

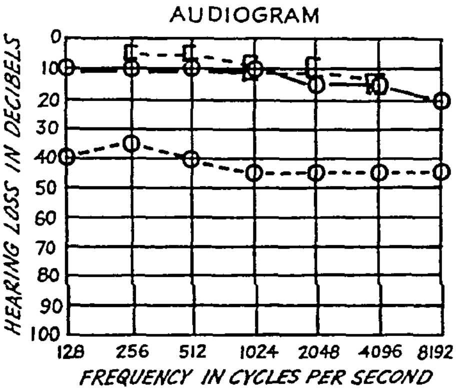
Eighteen cases are still under observation and have not been included in these results; they include some young children in whom a general anæsthetic was required and little co-operation in after care can be expected. Rather than repeat the anæsthetic, it may be wiser here to adopt preventive meatures until they are of an understanding age.

Illustrative Cases

1. HEARING IMPROVEMENT

H.S., æt. 32 years, had suffered from occasional earache and otorrhœa for many years and noticed slight deafness. Examination showed a clean dry central perforation of the right drum anterior to the handle of the malleus; she could hear a whisper at 3 ft.

After the second treatment the perforation healed, leaving an intact mobile drum with no visible scar, and she could hear a whisper at 10 ft. The audiogram shows this 30 db. rise.



This is evidence of the dramatic hearing improvement which may result as well as of the freedom from the risk of recurrent otorrhœa.

2 SOCIAL REHABILITATION

In some cases there may have been little obvious hearing loss and so no improvement there, but freedom from irksome restriction is reward enough

L A , æt 10 years, son of one of the Staff at the Swimming Baths, was learning to swim and at home in the water, he had every opportunity of access to the pool, but following an otitis media and persistent perforation, he suffered otorrhœa on bathing and after every cold, and swimming was forbidden Now, with his drum intact once more, he is swimming well and freely

3 MILITARY SERVICE

Persistent perforation renders a youth unfit for military service, although he may have no trouble and no apparent hearing defect, and this may be a great disappointment

J D , æt 19 years, had had occasional slight otorrhœa for two years, but his hearing was excellent, he was declined for service, but after closure of the perforation at the first attempt, was accepted

4 FOLLOWING MASTOIDECTOMY

In other cases, following the modified radical mastoid operation, there may remain a persistent tympanic perforation exposing the middle-ear mucosa

J McK had a right modified radical mastoid operation followed by complete epithelialization of the cavity, but leaving a clean dry central perforation of the drumhead Cautery and sponge-clot technique resulted in an intact mobile drum, while the audiogram showed some low-tone improvement

Summary

It would appear that by this conservative modification of the old trichloracetic acid cautery accessible tympanic perforations can be effectively and rapidly closed provided there is no gross deficiency, active chronic suppuration, or marginal destruction

Of 50 cases undertaken

36 (72 per cent) healed well
11 did not complete treatment
3 require surgery

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CLINICAL RECORDS

GANGLIONEUROMA OF THE LEFT MAXILLA AND ORBIT

By HUSSEIN H TOPPOZADA (Alexandria)

Introduction

THIS tumour, composed of adult ganglion cells is rarely seen It is much more common in the ganglia of the vegetative nervous system However, a few examples have been described in the brain, and the neighbourhood of the third ventricle appears to be their most favoured site It is interesting that many of the nuclei in this region constitute centres for the vegetative nervous system. In the cord, the neurinomata most frequently occur on the posterior nerve roots Encapsulated, they are found in the adrenals or sympathetic ganglia (Biggart, 1949).

In relation to otolaryngology it is rarer still Spiess, reports a pedunculated ganglion neuroma arising subglottically below the anterior commissure in a man 31 years of age Histologic picture showed malignant transformation He also refers to other known cases in this region

Axel Key, involving the ala nasi
Gaymuller, involving the lower mastoid area
Woods, involving the right parotid region
Benda, involving the parotid region
Glainsky, involving the deep cervical region
Freund, involving the deep cervical region
Summerfeldt, involving the submaxillary region
Eigler, originating from the posterior wall of the larynx (Spiess, 1929).

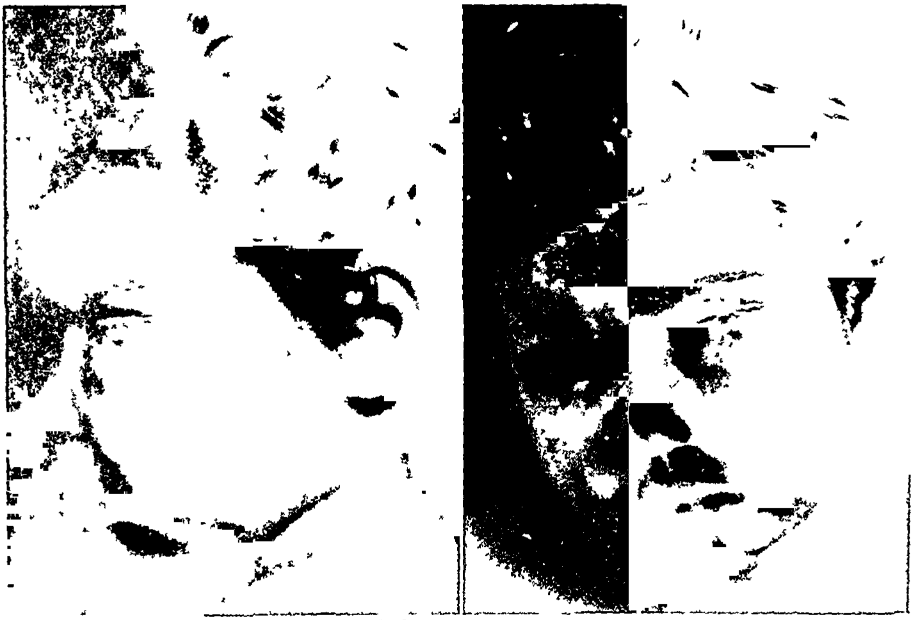
Key, 1879, gave an account of a ganglioneuroma of the nose in a man aged 31 years, presumed to have arisen from the infra orbital nerve (Willis, 1948a)

The following is another rare case

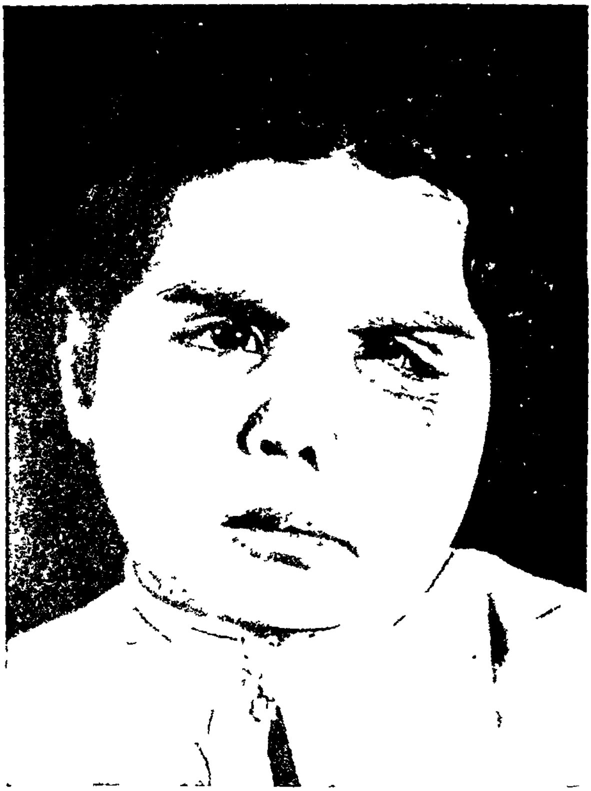
Case Report

A girl, aged 4 years, was brought to hospital in *November, 1950* The parents dated the history to 4 months before, when a small uniform bulge of the left side of the face was noticed, which gradually became more apparent until it reached the presenting condition, opposite the region of the left maxilla and lower eyelid and causing their forward bulging Diplopia started early in illness but no definite timing could be elicited Otherwise the girl was normal and in apparently good health

On examination, the left side of the face in the region of the left maxilla and lower eyelid was found to be bulged forwards by an underlying rounded,

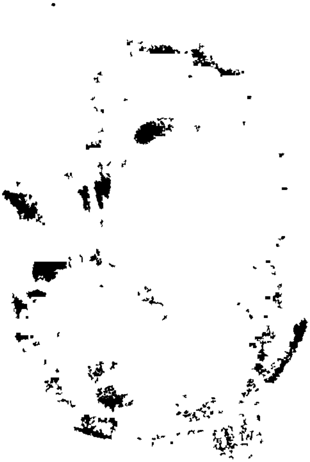


Before operation

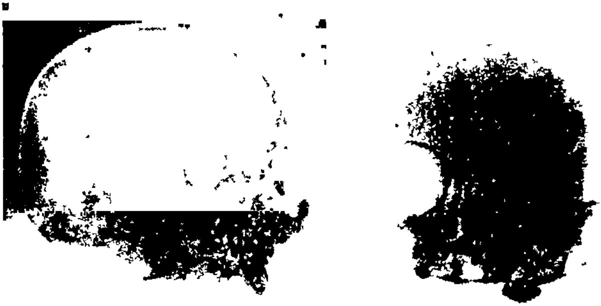


30 days after operation.

FIG. 1.



Occipito frontal view



Oblique view

Occipito mental view

FIG 2

Preoperative plain radiographs in various positions

Bone: Removed piece of left maxilla and zygoma in same position as in the body. Note the thickened infra-orbital ridge.

Tumour: the superficial ant. surface.

The lower surface adjacent to the Lt. maxilla and the intact hard palate.

The cut across deep part.

Remnant of tumour constituting the deep part which was removed in two pieces.

FIG. 3.

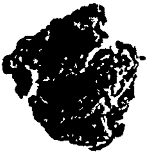
painless, diffuse swelling. The left palpebral fissure was narrowed and the lower eyelid everted, with slight upper entropion. The eyeball was pushed upwards and slightly forwards, but was free. The overlying skin was reddish but healthy. The hard palate and vestibule of the mouth were not affected.

The swelling was not hot nor tender. Its surface was smooth, uniform, with a hard consistency in the maxillary region but only firm in the left lower eyelid region. The overlying skin was not adherent but was tense. The upper part of the left maxilla and infra-orbital ridge was palpable but thickened. The swelling was fixed in its place.

The interior of both sides of the nose showed congested, reddish, inflamed mucous membrane of ordinary rhinitis. The left side showed no bulging of the lateral wall or disturbance of anatomical structure and no mass. There was no cervical lymph node enlargement.

Bone back surface of the removed piece of the Lt maxilla and zygoma partially covering the tumour

Tumour the upper surface adjacent and pushing the orbital contents



Remnant of tumour mass posterior view

Radiology Report "There is expansion of the left maxillary bone with areas of bone destruction as well as some reactionary new bone formation affecting nearly the whole of the left maxilla and extending upwards and laterally to the zygomatic bone and zygomatico-frontal articulation Picture is unlike an inflammatory lesion and is suggestive of a slowly growing tumour "

The Wasserman reaction was negative

Biopsy was taken through an incision of the left lower eyelid and revealed a "ganglioneuroma"

The treatment was surgical removal The operation was performed by Prof Aabdeen and myself The anæsthesia was ether and oxygen by the closed method introduced through an endotracheal tube placed through the mouth The nasopharynx was packed with ribbon gauze to prevent any blood from trickling to the larynx from the back of the nose

Novocaine 1 per cent., with adrenalin 1/10,000 solution was injected subcutaneously along the line of incision to lessen shock and hæmorrhage. A left paranasal incision was made and extended over the inferior orbital margin to its lateral end. The incision was carried down to the bone and the triangular flap of skin, subcutaneous tissue and periosteum, was undermined outwards and downwards. The anterior surface of the maxilla was fully exposed and found intact, forwardly expanded and thickened. A mass was found to occupy the following position:

Its upper surface was seen pushing the orbital contents upwards.

Its anterior surface was partially hidden by the thickened infra-orbital ridge and by the upper part of the anterior surface of the maxilla lower to the infra-orbital foramen.

This part was chiselled and removed, thus permitting a fuller exposure of the mass, which was found to be firm, smooth surfaced and covered with a capsule. A blunt dissector passed extracapsularly around revealed no adhesions to the surrounding tissues. The infra-orbital plate was not present. The lacrimal sac was retracted inwards. Extracapsular dissection was carried out far backwards and then the mass was cut through removing the already dissected part and allowing better exposure for dissection of the remaining part, which was removed in two pieces, and denoting a cone shape. No bleeding was encountered during the whole process. At the bottom of the cavity a soft tissue was felt denoting the pterygo-maxillary fossa contents. The hard palate and dental margin were found intact. The infra-orbital plate was not present so also for the infra-orbital nerve.

A rubber drain was put at the lateral end of the inferior orbital margin incision. Sulphonamide powder mixed with penicillin 100,000 units in 1 c.c. saline, was transformed to a paste and left in the cavity. The skin incision was stitched by interrupted fine silk worm gut. The skin edges were washed with saline solution containing adrenalin 1 : 10,000 by introducing the tip of a hypodermic needle mounted on the syringe in between the stitched skin edges, to wash off the blood clots and stop new clot formation. This procedure allows better healing with the smallest scar. The incision was covered with fine layers of cotton in collodion to spare repeated dressing.

Post-operatively, parenteral penicillin-streptomycin was given for 6 days. 150 c.c. saline solution and paraldehyde 15 c.c. were given per rectum, one hour after operation. The rubber drain was removed after 48 hours and its skin wound was stitched to complete closure. The stitches were removed on the seventh day. Albucid 10 per cent. drops were used four times daily to the eye and atropine ointment once daily kept the eye at rest. The patient received tonics and was kept under observation for one month and then discharged cured.

Pathology Report: Macroscopically: "A swelling, more or less oval in shape, measuring 3·5 × 2·5 × 2 cm. It is well circumscribed, encapsulated by delicate fibrous tissue which is not abnormally vascular. On section it is opaque white in colour, rather homogeneous in appearance and shows a few opaque yellowish specks. No marked vascularity."

Microscopically: "Reveals myelinated nerve fibres, with their axis cylinders running in various directions together with scattered large ganglion cells.

Besides, there is a fair amount of fibrosis Various constituents were proved by specific stains Blood vessels are well formed and not unduly increased Conclusion 'ganglioneuroma' "

Follow up In *March, 1955* the girl was recalled to hospital and re examined Some deformity of the cheek and side of the nose were found due to reactionary bone formation, resultant probably of the operative trauma inflicted on the periosteal covering of the region thus disturbing its physiological function, to check bone formation Clinically there was a depressed scar of the inferior orbital margin Bony expansion of the lateral nasal region, the alveolar border and the left zygoma Irregular teeth growth on the same side There were no changes in the hard palate The lateral wall of the left nasal cavity was bulging slightly medially, the inferior concha coming in contact with the nasal septum

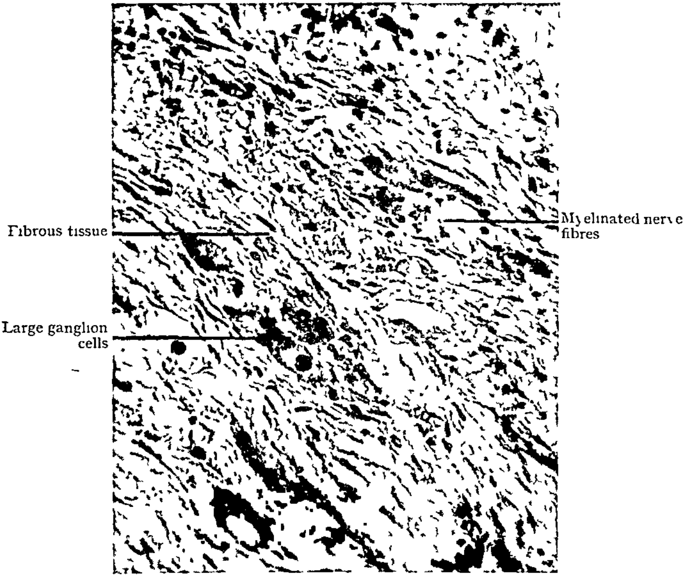


FIG 4

Microscopical appearance of the ganglioneuroma (× 120)

Radiologically "As compared to the previous X-ray films, the left maxilla is now nearly reformed, but showing more sclerosis It is slightly more thickened than the right side The left maxillary sinus is less well developed than the right, and its lower and lateral part is obliterated by the bone sclerosis seen "

No plastic procedures were found advisable for the present time due to the young age of the patient. Thus, the liability to later change and dissimilarity.

Comment

Regarding the embryological origin of the tumour, it is important to explain the presence of neural crest cells in the site of the tumour. They might owe their presence to:

(1) Migration from the semilunar ganglion or the sphenopalatine ganglion: It is gathered from different sources that migration of neural crest cells does occur. Provision for the ciliary ganglion is made early by the migration of cells from the major ganglion, along the developing ophthalmic division. Similar migrations along the divisions give rise to the sphenopalatine, otic, submaxillary ganglia (Gray, 1942).

There is no reason why similar cells do not occur along the infra-orbital nerve.

(2) Arise from the cells of the sheath of Schwann:

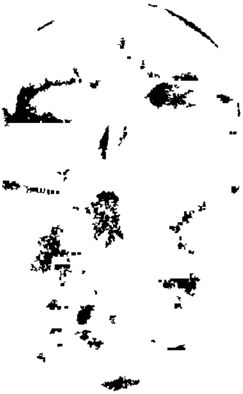
The peripheral nerve fibres are enveloped by a cellular sheath of Schwann. The component cells have two sources of origin. Some differentiate from the tissue of the early neural crest (Detwiler and Kehoe, 1939) and slightly later many others emerge from the neural tube by way of the ventral roots (Raven, 1937, Jones, 1939).

The infra-orbital nerve is medullated and its sheath of Schwann possesses cells of neural crest origin. These cells might preserve their earlier capacity to produce ganglion cells.

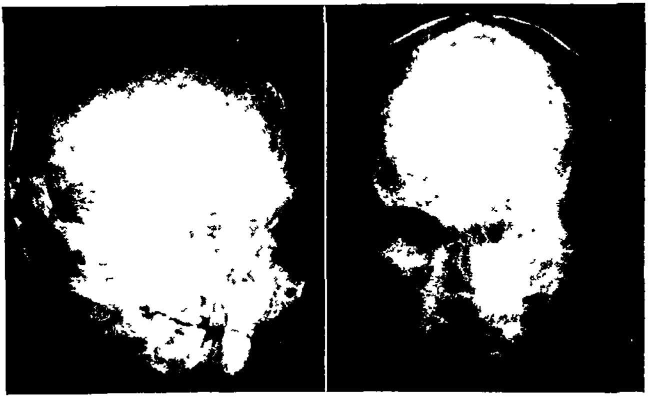
(3) Arise from the mesenchyme of the first arch:

In addition to forming the spinal and cerebral ganglion cells, certain neuroblasts of the ganglion crest are believed to migrate ventrally and differentiate into some of the cells of the sympathetic ganglia. Other crest cells become encapsulating and sheath cells, while still others mingle with the general mesenchyma (Stone, 1922). In man contributions to the first and second branchial arches is suspected (Bartelmez and Evans, 1926). The maxilla and zygoma are membrane bones derived from the mesoderm of the first branchial arch, which contained neural crest cells may give rise to the tumour.

Regarding the pathological aspect, this tumour is histologically benign and slow growing. It contains ganglion cells and glia cells, in short, it is a mature type of true neuroma (Lichtenstein, 1949). While many of them appear during childhood, in considerable proportion of cases their appearance is delayed until adolescence or adult life (Willis, 1948b). "Malignant ganglioneuromas" with metastasis in neighbouring lymph glands have been reported by Bencke, Brener and others. Their proliferation and metastasis must depend on the presence in them of immature neuroblastic cells. Fully differentiated ganglion cells do not multiply and hence a pure ganglioneuroma composed wholly of differentiated cells would be incapable of further growth or metastasis. The metastatizing elements of a tumour must be immature proliferating cells which may or may not attain recognizable nerve cell differentiation in their new sites (Willis, 1948b).



Occipito frontal view



Oblique view

Occipito mental view

FIG 5

Follow up plain radiographs 4½ years later

This tumour thus arising from the infra-orbital nerve and slowly growing, acted as a space occupying tumour causing displacement of the eyeball, bulging of the lower eyelid and cheek, and pressure destruction of the upper part of the maxilla, preventing the growth of the left maxillary sinus, of which no trace was found on radiography or at operation. The later development of the maxilla on the operated side within approximately the same size as the non-operated and considering the underdevelopment of the maxillary sinus, strongly supports Schaeffer's view that it is erroneous to think of the paranasal sinuses as forcing the facial parts to develop, or that the development of the sinuses is delayed until all of the parts have grown sufficiently to permit of their expansion (Schaeffer, 1919).

Acknowledgment

I wish to thank Prof. Aabdeen, head of our department, Prof. Assar, head of the Anatomy Department, Dr. M. L. Bayyoumi of the Pathology Department, Dr. G. Massoud of the Radiology Department for their very meticulous care and guidance in preparing this paper. Also, Mr. Maher of the photography Department, for his collaboration.

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TRACHEOSTOMY SUDDEN DEATH FROM DELAYED AIR EMBOLISM

By N ASHERSON

Air Embolism

Air embolism results from the aspiration of a small or large volume of air through any operative rent in the venous circulation It is liable to occur during any operation in the neck In the unique case recorded here, it occurred in the early post-operative period

The deep inspiratory movements of the chest produce a marked negative venous pressure in the veins of the neck, predisposing to the sudden indrawing of a large volume of air through any accidental tear, into the venous circulation Such a condition is apt to obtain in the presence of respiratory obstruction and the resultant extreme distension of the cervical veins

If the volume of air sucked into the vein exceeds 100 c c , sudden death will result (In the rabbit, an injection of 5 c c of air into a vein in the ear is used to kill the animal instantaneously) If the volume of air drawn in is smaller than 100 c c , definite clinical symptoms are produced If the volume is very little, no noticeable or detectable effects will be observed

The air enters the pulmonary circulation, the chambers of the right heart (where a large air lock is the cause of death), the pulmonary arteries and arterioles At autopsy, air bubbles are visible in the cerebral arteries, but not in the veins

Air Embolism following Tracheostomy

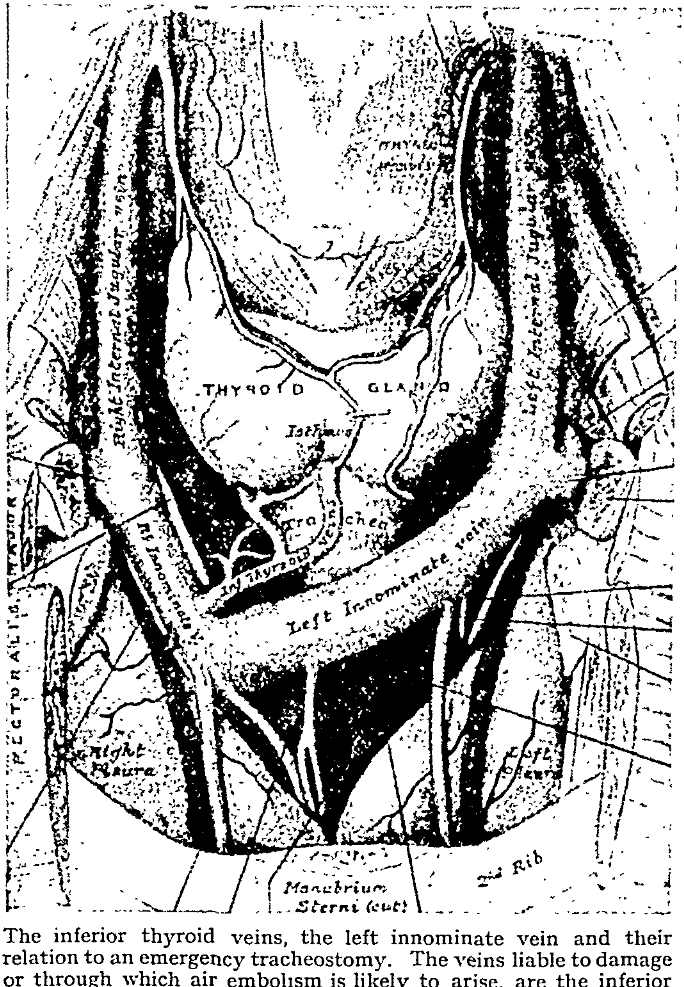
Death from air embolism, in the early post-operative period, after a successful tracheostomy, is an extremely rare sequel to this operation

The following case of such an unhappy incident is recorded as unless the cause of the sudden death had been ascertained at the autopsy, it might have remained obscure

The infant, aged 18 months sat up and suddenly dropped dead a few hours after a successful tracheostomy for acute laryngeal obstruction Autopsy revealed the cause to be air embolism (Another unusual cause—a bilateral pneumothorax—has already been recorded)

Tracheostomy for Acute Laryngeal Obstruction in an Infant, with Complete Relief, Sudden Unexpected Death a Few Hours After Operation Due to Delayed Air Embolism

An infant, aged 18 months, developed acute laryngeal obstruction, the inspiratory stridor being severe at rest It was completely relieved by a tracheostomy, performed under general anæsthesia, through an endo-tracheal tube The upper part of the trachea was exposed through a long midline incision, the isthmus of the thyroid gland being divided The veins of the neck were greatly engorged and the hæmorrhage was profuse, but completely controlled. The tracheal airway and bronchi were normal, dry and free from exudate. Towards the end of the operation, the infant coughed. This was followed by a profuse venous hæmorrhage from the lower part of the wound. It was controlled by gauze packing. The infant left the theatre in good condition, breathing freely through the tracheostomy tube but coughing occasionally. Four hours later, the child (who had been sedated with paraldehyde and whose condition hitherto was good) sat up and without any premonitory warning, suddenly dropped dead.



The inferior thyroid veins, the left innominate vein and their relation to an emergency tracheostomy. The veins liable to damage or through which air embolism is likely to arise, are the inferior thyroid or the left innominate.

(From Gray's Anatomy, 1944 by kind permission of the publishers, Messrs. Longmans, Green & Co., Ltd.).

At the autopsy, the laryngeal obstruction had been relieved completely:

there was congestion of the larynx and trachea The lungs were not collapsed there was no pneumothorax or any subcutaneous emphysema Death was due to air embolism The right chamber of the heart contained a great deal of air the arteries of the cortex of the brain were filled with air bubbles

Comment The coughing after tracheostomy must have dislodged a ligature from the plexus of the veins in the lower part of the incision, exposing a rent through which a large volume of air was drawn in suddenly into the venous circulation, as the infant took a deep inspiration prior to coughing

To avoid such a grim complication, all venous hæmorrhage, especially from the lower end of the incision, must be completely controlled A non-slipping ligature should be used, e g silk or thread

The difficulties and dangers of operations in the neck in the presence of great venous congestion have been described vividly by Hamilton Bailey ("Emergency Surgery")

"Under general anæsthesia, particularly when the airway is not entirely free, intra venous pressure rises diminutive veins wax proud Many tributaries become formidable vessels, the internal jugular vein and veins of kindred girth billow into mighty bags of blood that collapse suddenly on inspiration, almost at once to rise again and well might burst should the patient cough the surgical initiate should resolve that his index finger will reflexly enter the wound to plug a breach, at lightning speed to stem the hæmorrhagic deluge and guard against the very real danger of air embolism "

The Point of Entry of the Air into the Vein

The inferior thyroid veins, two in number, extend from the lower border of the isthmus of the thyroid in the midline and form a plexus in front of the trachea "From this plexus the left vein descends and joins the left innominate trunk, and the right vein passes obliquely downwards, and to the right to open into the right innominate vein, at its junction with the superior vena cava, frequently the two veins open by a common trunk in the latter situation" (Gray's *Anatomy*)

Doubtless, this plexus was the point of origin of the entry of the air

To avoid this tragic contretemps, the inferior thyroid vein should be seized horizontally by forceps, ligatured above and below and divided between

If the left innominate vein is situated in an abnormally high situation and is engorged, injury to it may be unavoidable

Tracheostomy above the thyroid isthmus would be the safer standard procedure

INFECTIOUS MONONUCLEOSIS PRESENTING AS MASSIVE TUMOUR OF NASOPHARYNX WITH CERVICAL ADENITIS

By M. E. N. SMITH (Swansea)

THIS case is presented for two reasons. Firstly, because no strictly similar case has been traced in the literature and secondly, because of the recent paper by Davidson, 1958, in which attention is focused on the nasopharynx in this disease, it being suggested that the nasopharynx participates in the clinical picture of infectious mononucleosis, particularly in its anginose form, to an extent hitherto unsuspected.

A girl of 15 was seen in Out-Patients and admitted therefrom to Hospital on *June 18th, 1957*. She was referred by her General Practitioner with a diagnosis of "sinusitis", having been quite severely ill for the previous two weeks. The sinuses had been X-rayed, for no known reason, at another hospital two years previously and the tonsils and adenoids had been removed, again for no known reason, at yet a third hospital, at the age of 6.

Clinical Findings

T. 102°

Massive glands, both sides of the neck, mostly in the posterior triangles. The over-all picture was strongly reminiscent of the bull-neck of diphtheria. Upon depressing the tongue with a spatula a striking picture was revealed. A large neoplastic-looking mass seemed to be occupying the whole nasopharynx as judged by the pushing forward of the soft palate and by the extension of the mass downwards a good ½ in. below the free border of the soft palate.

The nasal fossae were strikingly normal though the airways were grossly obstructed.

The nasopharyngeal mass was removed for biopsy with Irwin Moore forceps, under local anæsthesia, while the girl was still in the Out-Patient Department. The mass was friable and did not bleed unduly.

Investigations

X-ray. Sinuses normal.

Biopsy. "Hyperplastic lymphoid tissue with surface ulceration and acute infection. I think this must be a benign lymphoid overgrowth with superadded infection."

Blood (21.6.57). Hb. 77 per cent.; R.B.C. 3,790,000; W.B.C. 10,800; Differential—Polymorphs 36 per cent.; Lymphocytes 35 per cent.; Monocytes 29 per cent. Eosinophils Nil, Basophils Nil; Film—"Atypical mononuclears present indicating infectious mononucleosis."

Throat Swab. "Grew non-hæmolytic streptococci and *N. catarrhalis*. Both fully sensitive to all the antibiotics tested including penicillin. Diphtheroid bacilli also grew, resistant to penicillin but sensitive to all other antibiotics tested "

Paul Bunnell Reaction positive to titre of 1 in 448 Significant titre

Blood (*8 7 57*) Hb 93 per cent , W B C 10,000, Differential—Polymorphs 37 per cent , Lymphocytes 55 per cent , Monocytes 8 per cent , Eosinophils Nil, Basophils Nil Film—"A few atypical mononuclear cells are noted "

Discussion

Infectious mononucleosis was considered the probable diagnosis when the girl was first examined in the Out-Patient Department so that there was no delay in requesting a Paul Bunnell Other possible diagnoses such as sarcoma, reticulosis and antral neoplasm were, of course, entertained

However, in view of the high temperature, general ill health of the patient and absence of a precise diagnosis at that stage, a course of penicillin, 250,000 units, 6 hourly, for 5 days, was commenced immediately upon admission Apart from this, no treatment was given and none was indicated

The use of a dangerous and unpredictable drug such as Cortisone in an obviously self-limiting illness was not entertained for a moment

At no time was there the slightest suggestion of any need for tracheotomy In Davidson's case one cannot help wondering from the written account, if the precipitating factor were not the trauma of intubation for the laparotomy, particularly in so far as, apparently, the need for tracheotomy had not to be considered prior to anæsthesia and intubation However, Davidson's denial of this possibility and his own ingenious, physiological explanation of events is noted with interest

The nasopharyngeal tissue showed no hæmorrhagic tendency Custer and Smith (1948) record hæmorrhage from the nasopharynx as the cause of death in one of their nine fatal cases The writer has no experience of this disease as a fatal condition—but he is neither a physician nor a pathologist

In the present case recovery was complete and uneventful At the end of three weeks, the lymphoid mass in the nasopharynx had shrunk down into what looked suspiciously like adenoid tissue left behind at the previous T & A operation One wonders how the case would have appeared had the previous adenoidectomy been complete

Acknowledgments

I wish to thank Mr C P Robinson, whose case this was, for permission to publish, Dr O G Williams for the histological report and other members of the Pathological and Radiological Departments for their contributions

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SEQUESTRATION OF THE LABYRINTH

By J. F. JARVIS (Nairobi)

A MALE African child judged to be aged about 6 years was admitted to the Ear, Nose and Throat Wards of King George VI Hospital, Nairobi. No accurate history was obtainable, but he had a profuse purulent discharge from the right ear, a fluctuant post-auricular swelling and a complete facial palsy.

A sequestrum was seen presenting at the external meatus. At the operation, the sequestrum was grasped and on quite gentle traction the specimen shown in the photograph came away, the anterior end being the part which had presented at the meatus. When the mastoid was explored by a post-aural incision, a large smooth granulation-lined cavity was found occupying a large part of the petrous temporal bone. There was, of course, no trace of the facial nerve, nor was there any leak of cerebrospinal fluid into the wound. The granulations were not removed and this large cavity was drained and after a few days grafted with split skin. It healed soundly and rapidly with no further trouble.

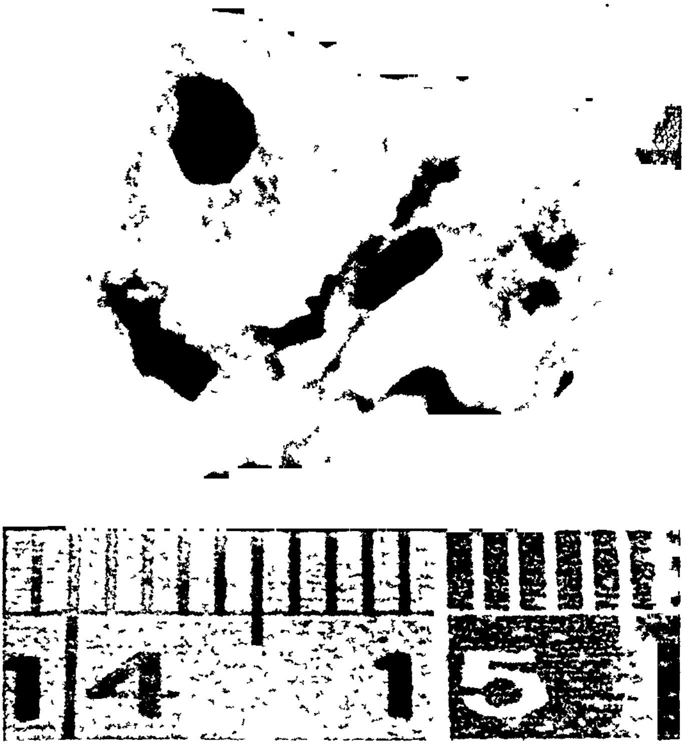


FIG 1
Sequestrum of labyrinth Scale in cm

The specimen which is preserved in the museum at King George VI Hospital shows the whole labyrinth intact and with only slight erosion of the dense bone of the canals In the photograph the three canals, promontory, oval and round windows and the facial canal can all be clearly seen

At some period during the formation of this sequestrum the meninges must have been breached and the great interest of the case is that at no time was there any history or evidence of meningitis It can only be assumed that a very effective process of obliteration of the subarachnoid space developed as an adequate barrier

A similar situation was found in another African child who presented with the whole frontal bone as a sequestrum, again with no meningitis and a complete recovery after removal Here infection was from a penetrating wound and not from the frontal sinus

A number of cases of gross sequestrum formation in the temporal bone have been seen at King George VI Hospital, due largely to the advanced stage to which ear diseases in the rural African population may be allowed to progress before treatment is sought When I showed the specimen to the late Mr W E Powell, he told me of a case of his where syringing of the ear had produced the whole cochlea which fell with a "ping" into the receiver

One of our adult cases was proved to be tuberculous in origin and besides gross destruction of the mastoid he had a sequestrum in the vicinity of the internal carotid artery Fearing that this might encircle the artery the temptation to remove it was resisted! It finally fragmented and came away spontaneously, and after prolonged anti tuberculous chemotherapy the cavity healed

SOCIETIES' PROCEEDINGS

ROYAL SOCIETY OF MEDICINE—SECTION OF OTOLOGY

President: I. SIMSON HALL, F.R.C.P.Ed., F.R.C.S.Ed.,

March 7th, 1958

SHORT PAPERS

CONGENITAL ATRESIA OF EXTERNAL AUDITORY MEATUS

By G. LIVINGSTONE (Oxford)

AT Oxford there is a large Plastic Surgery Unit. Mr. Eric Peet, who is in charge, was trained as an Ear, Nose and Throat Surgeon, but strayed from the fold to become a plastic surgeon. He is interested in the reconstruction of the outer ear in cases of congenital atresia and microtia, and it is only because many cases are referred to him that I can produce this series covering the last three years. Mr. Peet has done the plastic reconstruction, while I have only done the bone surgery and I feel that these operations should always be combined affairs.

In the last three years there have been a total of 16 cases, of which four were bilateral, and operations on 21 ears.

CONGENITAL ATRESIA

1955-1958

	Bilateral	Unilateral
Males	.. 2	9
Females ..	.. 2	3
	4	12

Total cases 16
Total ear operations 21

This is not the occasion to discuss the merits or demerits of plastic reconstruction of the pinna nor to produce a detailed paper on atresia, but this paper describes the techniques used at present at Oxford.

The reason why cases of atresia have potentially good hearing lies in the difference in the development of the outer and inner ear; the inner ear develops from an out-growth from the hind brain; while the pinna and external auditory canal come from the first branchial cleft. In the middle ear the malleus and incus come from the first branchial arch or Meckel's cartilage, while the stapes develops from the second branchial arch The styloid process and the lesser cornu of the hyoid bone are the other remnants of this arch

Usually when the pinna is malformed, the inner ear and stapes are normal, and it is in these cases that hearing may be established if a new meatus can be constructed to allow sound waves to reach the stapes

Bilateral cases are more of a problem to deal with than unilateral ones The question of when to operate is of fundamental importance as the development of speech and listening starts very early in life The hearing child begins to understand language and word discrimination from the age of fifteen months, possibly earlier, and at two and a half years he should be talking It is during this crucial time that the hearing centres are educated to word patterns, and unless a child is hearing spoken words during this period he will find it difficult to develop normal speech

To allow the child with bilateral atresia therefore to make the most of his residual hearing during this important phase of language development, early operation is advised One ear should be operated on between eighteen months and two years of age and the second one a few months later if normal intonation does not develop In every case auditory training should be started as soon as possible, and a hearing aid supplied

The disadvantage of operating at the earlier age is that very young children are unco operative, and an audiogram cannot be obtained

In unilateral cases the decision of whether or not to operate can be left until later The operation is justified partly on psychological grounds, and partly because there is a good chance that the ear concerned may be made efficient The age of choice in which to operate is between 6 and 8 years, in order to allow time for the plastic surgeon to rebuild the outer ear before serious schooling starts

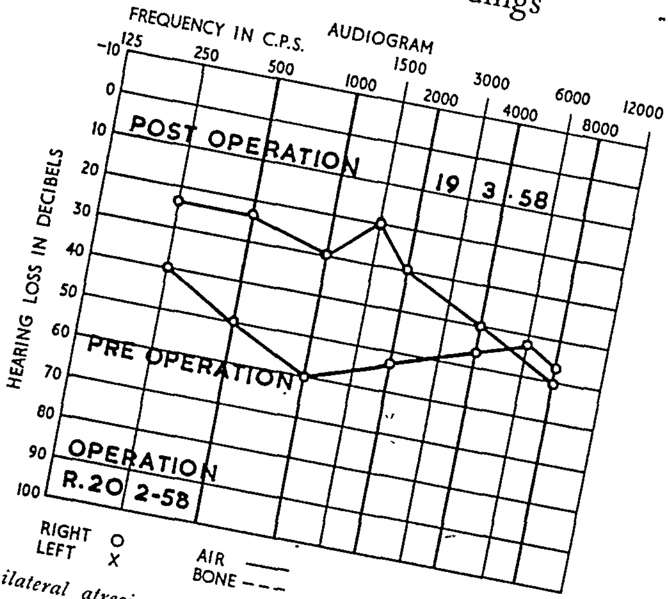
Turning now to technicalities X-rays were found of little value beyond demonstrating whether there is a cellular or dense mastoid An X-ray showing no ossicles is no reason for not exploring the middle ear cleft

In 16 cases, 12 mastoids were dense although in most of these a middle-ear cleft was present The incus and malleus are frequently fused into a solid mass, raththe surgical approach to them is much easier when the mastoids are cellular and er than dense

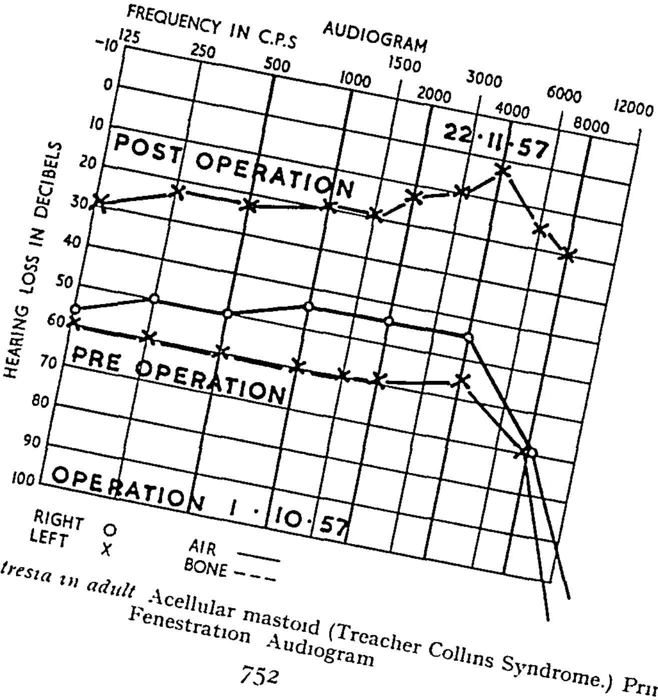
The text-books and various monographs stress the possibility of finding an aberrant facial nerve and the dangers to it of undercutting the skin round the incision So far, no abnormal positioning of the nerve has been found in any of the cases in this series, although frequently the nerve is never seen

Sometimes a dimple shows in the skin where the external auditory meatus should be, but usually there is no indication, and the shape of the incision is left to the plastic surgeon who decides in each case the tissue he wishes to preserve Consequently the incision is often an irregular zig-zag, although whatever shape it is, by the time a retractor has stretched the wound to make the exposure adequate, the result will be the same The extensive perimeter of the incision is one reason why stenosis has not occurred in these cases

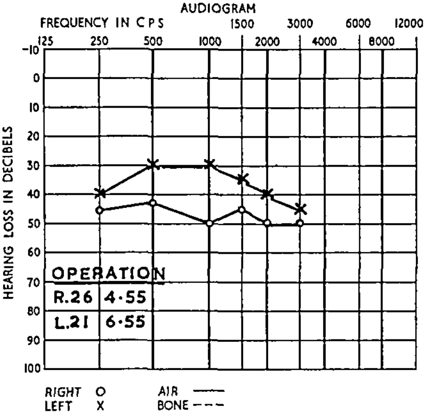
In the first of the bilateral cases the incision was shaped to include a pedicle flap of skin hinged at the back This flap was used to fold over the new posterior meatal wall, with the tip pressing down on the head of the stapes and the rest



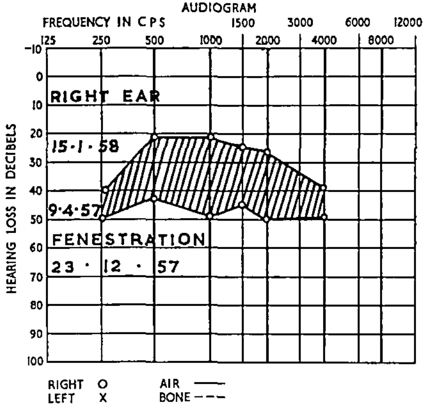
Case I Unilateral atresia in child Cellular mastoid. Tympanoplasty type III. Columella reconstruction Audiogram



Case II. Bilateral atresia in adult Acellular mastoid (Treacher Collins Syndrome.) Primary Fenestration Audiogram



Case III. Bilateral atresia in child First operation—tympanoplasty type III did not obtain adequate hearing in right ear
Audiogram



Second operation—fenestration external canal Audiogram closing the opening into the middle ear; the anterior wall was lined with a split thickness skin graft. This technique is not now used in this Department.

Once the ossicles are exposed, magnification is essential, and a dissecting microscope with adequate focal distance should always be used.

The best method to approach the middle ear is a problem, but if difficulties are encountered Pattee's method is a useful guide. This entails judging where the meatus should be in relation to the tip of the mastoid and the condyle of the jaw, and burring at this point to expose the middle fossa dura, then tracing the bone down to the tegmen and then backwards to expose the lateral sinus and working down the sino-dural angle to reach the external semi-circular canal and the opening into the middle-ear cleft.

The large meatus is not necessary as stenosis has not occurred, and the surface of the bony meatus is not polished, as the graft is supposed to take better on rough bone.

When the antrum is reached the problem arises of what to do in the middle ear. Ombredanne, in Paris, advocates an immediate fenestration of the external semi-circular canal, but this has not been done in this Department. If fenestration on a baby goes wrong, the labyrinth is destroyed and no further operation will help. Cases have, however, been revised, and a fenestration done when the hearing improvement was not up to practical level, possibly due to scarring in the middle ear, and where the bone conduction audiogram was normal.

Instead of a fenestration in cases where the fused malleus and incus are freely mobile, the graft is applied directly to the ossicles. When, however, the bones are fixed, the malformed incus is removed, and after the middle-ear cavity has been defined and the stapes exposed, a tympanoplasty type III is done.

In these cases, once the fused mass of incus and malleus is removed, the stapes is usually found in some shape or form and the setting is ideal for a tympanoplasty type III.

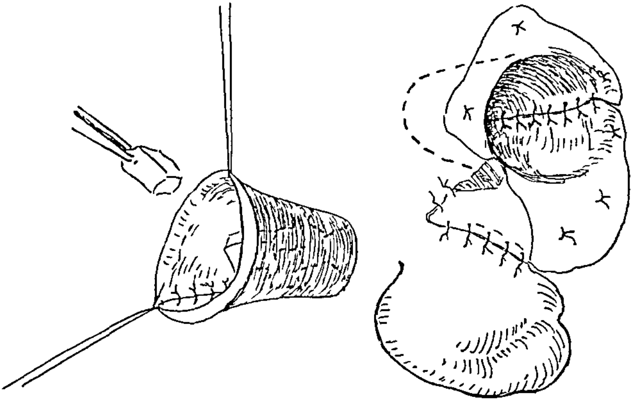
Great care is taken to approximate the new graft on to the head of the stapes leaving the rest of the middle-ear cavity severely alone. In this way a columella effect is obtained, sound being transmitted through the graft and stapes to the oval window.

In the early cases of this series a stent mould was used to hold the graft in position, but this method has now been abandoned. At present nothing hard is used as plugging in the cavities, but the newly-formed meatus is lined with split thickness skin graft taken from the thigh. This graft is sewn into the shape of a bag with the raw surfaces together and then turned inside out. The bag is stitched to the skin edges of the incision. It is then packed down with pledgets of plastic sponge soaked in Neomycin cream so that it approximates on to the head of the stapes and on to the bony walls of the cavity. Gelatine foam is packed around the stapes and eustachian tube so that this bag will not press into the middle ear. This graft has taken *completely* in all of the 19 ears in this series.

After the operation the ear is not touched for 10 days, when the stitches and plastic sponge are removed, and replaced with pledgets of cotton wool soaked in liquid paraffin. This is repeated weekly for a further 3 weeks, then a mould is taken for fitting a hearing aid. When possible an audiogram is also done, but usually improvement in hearing has been noticed after the first dressing.

In the young children pre operative audiograms could not be taken, and only the audiograms taken a year and a half after operation can be shown But during that time the children had learned to talk and they now have normal intonation

The Treacher Collins type of cases are the most difficult, and in three in which fenestration was attempted, in only one was a canal found, but with a fenestration the hearing result is at present excellent The other two have some improvement due to the middle ear cleft being opened



In conclusion the following points should be stressed

(1) Bilateral cases should always be operated on, and the age for operation in these cases should be between eighteen months and two years

(2) Unilateral cases should also be operated upon but at a later age, either when the patient requests it, or when the plastic reconstruction demands the external auditory canal

(3) The operation of choice in children is a tympanoplasty type III If this does not produce adequate hearing, a fenestration can be done later But it is not advised unless it is confirmed that one window does not move

(4) At present a Thiersch graft bag reconstruction of the meatus is the operation of choice

(5) The dangers of stenosis and injury to the facial nerve are exaggerated.

SURGERY OF CONGENITAL ATRESIA OF EXTERNAL AUDITORY MEATUS

By H. S. SHARP (London)

SINCE 1947 I have seen some thirteen cases of congenital atresia of the meatus. Ten of these have been unilateral and three bilateral. On eight I have already operated, and others are awaiting operation.

The pinna in every instance has been more or less abnormal, and most, though not all, have needed plastic surgery or prosthesis. In the majority of cases there has been no other congenital deformity. In the Treacher Collins syndrome the facial abnormalities rather over-shadow the aural, and in these cases I do not consider the repair of the auditory meatus on cosmetic reasons as important as in the more common uncomplicated aural malformation only.

The indications for operation in my opinion are two—cosmetic and functional. In the unilateral cases, the cosmetic is the more important. It is surprising how unkind children can be to a fellow scholar without a meatus—it seems to attract more attention than a malformed pinna.

In the bilateral cases the functional reason becomes important, because of the deafness.

The best age to perform the operation is between 4 and 5 years of age, so that the child can be ready for school. My experience of doing the operation earlier has been rather disappointing, as the field is that much smaller, and the new meatus more liable to closure. In bilateral cases the child should be given a hearing aid until 4 to 5 years old.

As regards preliminary investigations I do not now do any X-rays. The information as regards canal system or ossicular formation rarely gave me much help in the early cases, in which full X-rays were done.

I tried fenestrating the lateral canal in the first few cases I did, but was not convinced that the fenestration was playing any part in the resulting improvement in hearing, and in two cases I found no proper membranous canal, so I have given this up. Tympanoplasty I have not considered, because of the gross abnormality of the middle ear in all the cases. The ossicles have nearly always consisted of a malformed incus ankylosed to the head of a rudimentary malleus.

The technique I am using at present is as follows:

A post-aural incision is used. The pinna may bear no relation to the bony mastoid cortex, so the mastoid tip must be palpated before the incision is made, and the cut made accordingly. There is no place for any sort of endaural approach in this type of case.

As far as the plastic surgeon is concerned with the repair of the pinna, the incision should be made as far back as possible within reason.

The incision is then deepened to the mastoid cortex. The ring retractor is used and I find it by far the best for all forms of post-aural mastoid surgery.

The bony work is now completed, the drill and burr have to be used to ensure a smooth cavity with no ridges or crevices to facilitate the fitting of the skin graft. Incidentally in only one of these cases have I found any abnormality in the anatomy of the facial nerve.

The pinna is now turned back, and the incision for the new meatus made.

It is almost impossible to make the opening too big the pitfall to avoid is making it too small There is an anterior hinge on this outline of the incision

The flap resulting from the new meatal opening is then turned forward and thinned down to full thickness skin depth, and it is eventually turned inwards to form the anterior wall of the new meatus

The next step is to turn back to the mastoid cavity and apply a skin graft to it I use black nylon gauze, such as one finds in ladies' stockings, as a mounting for the graft This is lightly spread with vaseline and holds the thin free graft well, making it easy to handle I find this nylon gauze far better than tulle gras, or linen or nylon fabric The graft for the mastoid cavity is cut with a Humly knife from the inner side of the thigh I use a piece of portex tubing in the new meatus at the conclusion of the operation

It is very important to get the graft fitting snugly Smaller pieces of graft are used to fill up gaps as necessary, and a gauze pack is then applied to keep it in position

There are two portex tubes used, the outer one is split throughout its length, and inside this is a smaller tube exerting gentle pressure on the outer one, and so to the graft lining the new meatus The anterior hinge full thickness skin flap is in position and the free graft is reflected from the rest of the circumference of the portex tube

This completes the operation One week later the pack is removed, with all the nylon mesh The two portex tubes are reinserted, and a cage put over the ear The cavity is gently mopped out from day to day, and a week later an insert mould is made to fit the new meatus, in place of the portex tubes

It is very important to watch each case for at least six months, and I like them to wear the insert, which may need modifying from time to time, for three months

In my experience there is a great tendency for a slow stenosis to occur up to six months

Mr David Bawtree said, two cases of congenital atresia, both adolescent girls, had recently come under his care

On one he had found no external meatus and no recognizable ossicles, and had therefore carried out fenestration with Thiersch Graft with 20 db improvement in hearing

The other case had no external meatus but a normal ossicular chain Tympanoplasty with whole skin graft to cover the malleo-incudal joint area was carried out with Thiersch graft to the cavity A very satisfactory 25 db improvement in hearing occurred, but recurring stenosis had been found a problem and a further Thiersch graft was found necessary

What was the general feeling in regard to "Whole" or "Thiersch" grafts in such cases?

Societies' Proceedings

ENDAURAL SURGERY OF THE JAW JOINT

By A. S. DAVIDSON (Kettering)

I FEEL that some word of explanation is necessary with regard to the topic of my short paper.

It may well be asked what business the otologist has with the jaw joint. Briefly the answer might be that the surgical treatment of this area is not as yet, rigidly associated with any particular department, and in different hospitals either the orthopædic surgeon, the general surgeon, the faciomaxillary surgeon or even all three have these cases referred to them. In many instances there is a lack of a standard procedure, and sometimes there is reluctance to undertake operation in cases which have not responded to other measures.

It was in fact an orthopædic colleague who first stimulated my interest in this problem. He had a patient, a women of fifty, who for twenty years had complained of painful chewing with recurring episodes of locking of her jaw joint. These had become so frequent that in one month she had attended the Casualty Dept. on fifteen occasions, whilst on three of these occasions she required a general anæsthetic for reduction of the subluxation. Apart from these occasions there were many times when she successfully manipulated her own jaw, so that she lived in perpetual apprehension and pleaded for an operation.

My colleague observed that the technical difficulties of jaw joint surgery closely resembled those encountered in the mastoid, because in both instances one operated in a deep, dark, and frequently bleeding cavity, with some risk to the facial nerve or its branches.

Consideration of the incisions usually employed showed that the joint is commonly approached from its lateral aspect. After a horizontal, vertical, or T shaped skin incision the overlying soft tissues are divided until the joint is identified. Troublesome bleeding in this highly vascular area is common, and there is a risk of damaging the upper filaments of the facial nerve. An additional disadvantage is that the access afforded is very limited because the depth of the joint in a direction perpendicular to the skin surface is at least three times its extent in the antero-posterior dimension, as can be seen in the illustration which also shows that approximately the outer third of the posterior boundary of the joint is cartilaginous meatus, the inner two-thirds being the bone of the tympanic plate. Taking advantage of this fact, and using an "anterior" endaural incision, the jaw joint may be exposed for almost half its depth in addition to its lateral aspect, and the difficulties of a deep dark cavity thus minimized. An additional advantage lies in the fact that there is no danger to the branches of the facial nerve. As regards the cosmetic effect, the scar post-operatively is almost indiscernible.

The Anterior Endaural Incision. The position and shape of the incision are indicated by the plasticine placed on the dried skull (Fig. 1).

Advantage is taken of the fact that there is in the pinna a fibrous cleft, deficient of cartilage, lying between the tragus and the helix. An incision through this cleft allows the otherwise intact cartilaginous ring of the pinna to be opened out and mobilized, without damage to its integral structure.

For descriptive purposes, the incision is seen to consist of three parts: (1) A facial portion, about an inch long above the level of the zygoma and running downwards and backwards to the inter-cartilaginous cleft (Fig. 2), where it becomes continuous with (2) a meatal portion, also in the inter-cartilaginous cleft. This extends inwards for about half an inch along the roof of the meatus from the surface to the lateral border of the bony meatus. On the skull this corresponds to the inferior surface of the post-glenoid tubercle. (3) From this point the incision continues in the sagittal plane, extending round the anterior half of the meatal circumference at the junction of bone and cartilage.

Otologists will recognize this incision as a modification of that used for endaural mastoidectomy. The first and second portions are practically identical with those used in endaural mastoidectomy, whilst the third portion of the incision involves the anterior meatal wall instead of the posterior wall.

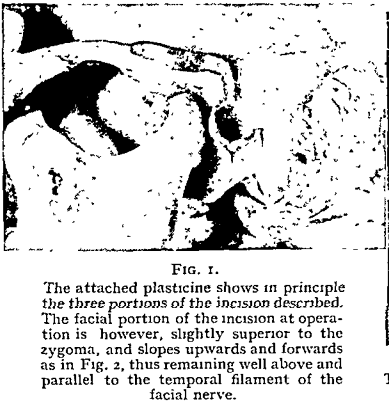


FIG. 1.
The attached plasticine shows in principle the three portions of the incision described. The facial portion of the incision at operation is however, slightly superior to the zygoma, and slopes upwards and forwards as in Fig. 2, thus remaining well above and parallel to the temporal filament of the facial nerve.

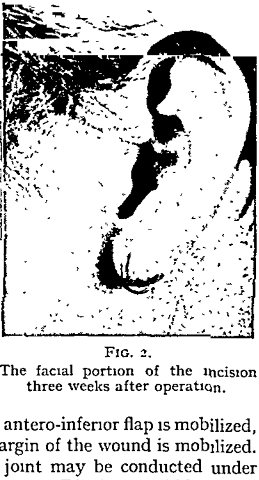


FIG. 2.
The facial portion of the incision three weeks after operation.

In the endaural exposure of the jaw joint, the antero-inferior flap is mobilized, whilst in mastoidectomy the postero-superior margin of the wound is mobilized.

Operation. Although operations on the jaw joint may be conducted under local anaesthesia with some advantages to the surgeon (Dingham and Moorman, 1951), it seems preferable to give the patients the benefits of general anæsthesia. Endotracheal intubation is desirable, as the patency of the airway may then be safely assured however much the head is moved. It is a point of some practical importance that the endotracheal tube should be passed, not through the mouth, but through the nose if this is possible without causing trauma. Routine oral intubation leaves the mouth in an open position with the condyle correspondingly moved forward in the wound. Conversely, the anæsthetist can help to deliver the condyle into the field of operation by firmly approximating the incisors when required

Hæmostasis. The area of the incision is infiltrated with a 1:100,000 solution of adrenaline; any local anæsthetic solution containing a vasoconstrictor will be equally suitable.

Exposure. The incision is made in the order described. Its first part begins about an inch anterior to and terminates at the cleft between the tragus and the helix. By incising skin and subcutaneous tissue only, the superficial temporal vessels ascending across the incision anteriorly are identified. These are ligated and divided, and the incision is deepened until the temporal fascia is exposed. An assistant retracts the lips of the incision, which is now extended deeply between the tragus and the helix along the meatal roof, the knife being carried right down to bone throughout. The inter-cartilaginous cleft can now be widely opened by retractors, and with a small-bladed knife the junction of the cartilaginous with the bony meatus is divided in the anterior meatal wall immediately behind the joint. To accomplish this separation the incision need be no more than one or two millimetres deep.

It will be noted that the primary incision extends deeply to bone only in its mid-portion, that is, along the inferior surface of the post-glenoid tubercle. Elevation of the soft tissues so as to expose the jaw joint is best started from this point. Beginning at this bony landmark and proceeding anteriorly, the periosteum of the zygoma is stripped forwards and downwards as far as the articular eminence. Thus the lateral surface of the joint capsule is exposed. The posterior portion of the capsule is now put under tension by the operator or assistant grasping the mandible through the towel and "overclosing" the bite. Beginning again at the post-glenoid tubercle, the meatal soft tissues are elevated from the posterior aspect of the joint and reflected downwards and outwards. A self-retaining retractor of the mastoid type is now inserted. It takes its purchase on the undisturbed postero-superior lip of the wound, and draws down the antero-inferior flap to expose the joint capsule in its lateral and postero-lateral aspects. A horizontal incision is now made through the joint capsule parallel to the lower border of the exposed zygomatic arch on to the condyle, thus opening the lower joint cavity only. The neck of the condyle is exposed by converting this horizontal incision into a T-shaped incision, when the bony insertion of the lateral pterygoid muscle anteriorly comes into view. This insertion is easily stripped with a periosteal elevator, and the condyle and neck are fully exposed by elevators placed anteriorly and posteriorly to the neck (Fig. 3). When the object of the operation is condylectomy, the condyle is then removed, using bone forceps, or other means. Interposition of soft tissue over the raw bone to prevent an ankylosis does not seem necessary, as it is separated from the temporal articular surface by the intact meniscus. For meniscectomy, the condyle is distracted exposing the meniscus, and an incision made into the superior joint cavity by dividing the capsule attached to the upper lateral edge of the meniscus (Fig. 4). This free edge is now grasped between forceps and its peripheral attachments are cut in succession, until the cartilage is freed entirely. Care must be taken not to damage the intact of mandible or glenoid. The wound is now carefully sutured, in the reverse order of its formation, that is, beginning at the meatal floor. Ease of access is gained by inserting all the necessary meatal sutures before any of them are tied. The meatus is now lightly packed, and a pressure dressing applied.

Post-operatively, patients are encouraged to masticate normally and to become ambulatory as soon as they wish. The dressing requires no special attention, apart from changing the meatal wick should the wound ooze. This slight ooze is more noticeable after condylectomy than after meniscectomy, owing to the "dead space" present in the former operation In no case has it been troublesome Sutures are removed in seven days

Most patients suffering from internal derangements of the jaw joint obtain relief from conservative measures directed towards correcting faulty occlusion and the resting of the joint Only a relatively small proportion of patients require surgical treatment, and in these cases the approach described above has been tried out since May 1952 in five cases for condylectomy, in ten instances for meniscectomy, in one case of bilateral arthroplasty for habitual dislocation in an epileptic, and also in the case of a patient who had suffered from bilateral bony ankolsis for fourteen years

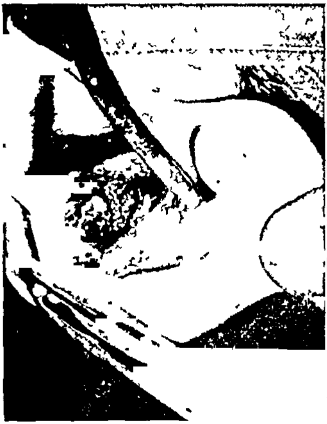


FIG 3

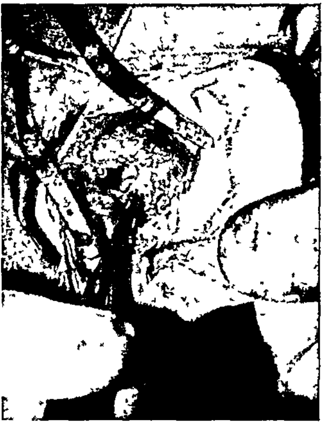


FIG 4

Fig 3 —The left condyle exposed in this patient the condyle shows areas of erosion The anterior border of the deep meatus is just visible in front of the posterior limb of the self retaining retractor

Fig 4 —Following removal of the condyle the meniscus was freed along its lateral margin superiorly and retracted downwards by forceps as shown so as to give an impression of the depth of the jaw joint

So far there has been no suggestion of meatal stenosis in any of these cases, in fact, the meatus is widened anteriorly following condylectomy on account of a slight hollow occupying the site of the former condyle

I gratefully acknowledge my indebtedness to my colleagues, W S Matheson, F D S R C S , and T K Davidson, M B , Ch B , D Phys Med (Eng), for their whole hearted interest and co operation in the treatment of these patients I am grateful to Mr R R Popham, F I M L T , for the photography

REFERENCE

DAVIDSON A S (1955) *Brit J plast Surg* 8 64

Societies' Proceedings

TETANUS FROM A MARINE SPONGE

By LIONEL TAYLOR (London)

A PATIENT aged 30 had otosclerosis. After trial of hearing aid she wished to try the fenestration operation in her left ear which was her worse ear.

A left fenestration operation was performed on *April 19th*. On *April 24th* she developed trismus and was seen by the dental surgeon who could find no dental cause. On *April 25th* the ear was redressed under general anæsthetic, but in the evening the trismus was worse. An aural swab was taken and put up. I/M ATS was administered (30,000 units) in addition to penicillin.

On *May 1st* trismus was a little improved, and the patient's general condition was much better. Organisms morphologically similar to *Cl. tetani* were isolated. On *May 6th* trismus was still marked and the next day bacteriological confirmation of *Cl. welchii* and *Cl. tetani* was obtained by culture. Large doses of intravenous ATS were given after due testing. Myanesia was started on *May 8th*, but trismus continued in varying degrees of severity and there were signs of generalized spread such as spasm of recti abdominis muscles and increased knee and ankle jerks. The ear was explored on *May 16th* to remove any dead tissue in view of the persistence of *Cl. tetani* in swabs, but only a healthy cavity was found. On *May 20th*, there was some twitching of left leg and right shoulder. Her condition steadily improved and she was discharged fit on *June 27th*, with some improvement in her hearing!

On *April 26th*, two marine sponges similar to those used at operation were sent for culture and they both yielded *Cl. tetani*. The dust in the operating theatre on window ledges and scialytic lamp was examined and also found to contain *Cl. tetani* and *Cl. welchii*. Although this latter may be considered a possible source of infection it seems more probable that the marine sponge used was the infecting agent. This case presented for interest because I believe this is not an isolated instance. The interest will now mainly be academic because the use of marine sponge has largely been replaced by spontex or other similar artificial sponges.

HEARING PROBLEMS IN A BABY

By NORMAN L. CRABTREE (Birmingham)

THIS case demonstrates many of the difficulties which beset us in assessing hearing function in the very young. Now that we are realizing the benefits which result from the alleviation of a congenital deafness by the use of a hearing aid to reduce the degree of the disability, and by the training of the parent and the child, we have become progressively concerned with attempting to obtain an assessment of hearing function at a much earlier age.

Our patient is a girl called Alexa, who is now 20 months old, who demonstrates many of the difficulties which we meet. She was an illegitimate child, actually of a Nurse, born in June, 1956. Apart from some trouble with fibroids during pregnancy, her ante- and post-natal history was normal. She went to another City for adoption, and when she was 5 months old, it was found that while she was apparently a child of normal intelligence and active in her interest of everything she could see about her she made little or no response to sound She was seen by a colleague who reported that she responded only to very loud sounds and he felt that she was extremely deaf The adoption was rejected Alexa returned to her mother in Birmingham and was fostered out with friends who fortunately had a child of their own some three months older It was under these circumstances that she first came to us

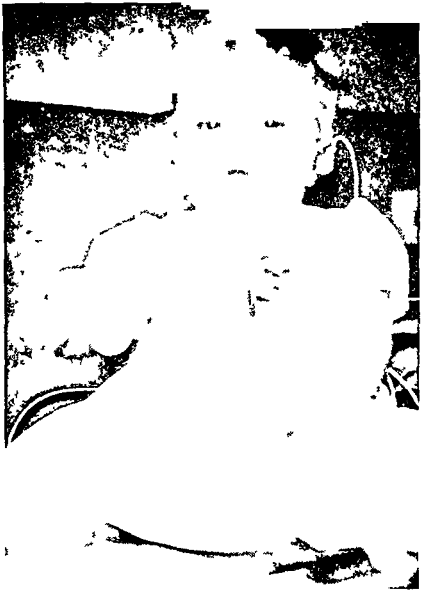


FIG 1

At the age of 6 months two weeks after the fitting Alexa contentedly wears her aid throughout waking hours

Our examination at the age of 6 months showed precisely the same findings as I mentioned earlier She seemed a bright intelligent and happy child but she made virtually no response to sound She was not interested in any of the stimuli which we presented to her and the only positive response we could obtain was a slight flicker of the eyelids to a very loud sound of the xylophone and drum near the ear

We agreed with the earlier opinion that she was extremely deaf We fitted her with a hearing aid in the left ear which she accepted easily and after two weeks was able to wear it all day

She has attended our clinic periodically since this, but we have, in fact, only seen her on seven occasions, during which a certain amount of guidance and help has been offered, but in the main, her progress has been dictated by the extremely sensible and understanding actions of the foster parents, and undoubtedly the good home conditions and the presence of the other slightly older child were greatly beneficial.

We found, after a few weeks, that she had already become interested in practically all sorts of sound, and by the time she was a year old, she responded to her name, rather to our surprise, at a distance of 12 feet in a normal conversation voice, without wearing her hearing aid.

Two months later we were able to obtain responses in the left ear to a variety of hearing tests up to a distance of eight feet, but we were not getting any response in the right ear. Three months later, when she was 17 months old, we found that she had developed a very positive response to hearing in both ears, and more than that, she was able to locate the source of the sound, and in fact, responded to and located sounds up to a distance of about 10 feet.

At this point, we took a film, which I would like to show you. The film has not been cut in any way. It is presented just as she was submitted to test, with the hearing aid disconnected, and you will see that the generally accepted test procedure is followed. We have an Audiometrician interesting her in toys at a small table, while behind Alexa is another Assistant, in this case a Teacher of the Deaf, who is distracting her by means of sounds which we found interested her. For the purpose of the film, the tests are carried out quite close to her, but in practice, we obtained the same response at a distance of 8 to 10 feet. You will see that we vary the tests frequently. Having obtained or failed to obtain a response, we change the sound source and return to it again, trying to produce a variety of stimuli which will continue to attract her interest.

You will see that we have the three varieties of response—the completely negative response, which results either from the child being too absorbed in her play, or alternatively not interested in the type of sound which is being presented, and would also follow from a failure to hear. Then the two varieties of positive response—the first of the attitude of listening or attention, and secondly the direct positive response of turning towards the source of the sound.

You will see that we are not above bribery with sweets during this test!

During this time her speech was developing. At the age of one she was already trying to say "Hello Mummy" in her own way, and this vocabulary has steadily increased.

I have a short recording of her present voice, which shows that it is of normal quality and timbre, and that the development of her vocabulary is probably not very short of normal for an intelligent girl of her age. Before I play this recording, I must ask your indulgence, because you will probably agree with me, that it is extremely difficult to get a child to perform when you want it to, and also we did not want to cook up anything extra special, but rather to leave it to the foster mother and Alexa herself to talk to you just as they would talk when they came to visit the Clinic. In fact, you are hearing just exactly what you would hear in the Clinic.

I would like to say a word or two about the machine, which falls rather short of a good tape recorder in its quality, but has the very considerable advantage that recording and reproduction is extremely easy to carry out in a Clinic The lack of the need to wind back the tape and the solidity of the record, makes it simple and quick to operate, and we are finding it very useful in our Clinic to record the sounds of the child and parent, and other sounds which may interest the child, and then to play them back, first at the original intensity, and then at louder intensities We find that, with a child who is partially deaf, we often get a helpful positive reaction from carrying out a test of this type, and it can be done in the course of a normal interview without any great complication

To summarize the case history of Alexa—we were faced with a child who, at 6 months of age appeared to be almost totally deaf, and with a change to a happy home environment with another child and helpful foster parents, and an atmosphere which one can only adequately describe as that of "mother love", the child has received much attention, has been talked to a great deal and her progress has really been that of a normal child Now, at the age of 1½, she appears to have normal or near normal hearing It does now appear that the particular early post-natal life to which she was subjected, and a lack of understanding of her needs on the part of the couple who originally adopted her but failed to treat her naturally, led her to become disinterested in sound, and already at the age of 6 months, she was showing herself as a child deprived of communication by hearing and speech

Now we have reached the point at which it seems pretty certain that Alexa's hearing is either normal or very nearly so it is interesting to speculate what would have happened had we removed the factor of deprivation and placed her in her present surroundings, but without a hearing aid Would she have progressed in the same way, or did the use of the hearing aid, by bringing sound to her more loudly, help her to develop an interest, and to achieve the present attainment level so quickly? Her response to sound in the left ear before she responded in the right, suggests this is probably the case

Another interesting point is that, within a few days of fitting her with her hearing aid, which was a Multitone 'Minuet she happily tolerated an intensity level of '6', and wore it at that level all day long She does, therefore, demonstrate that a baby will readily accept a hearing aid, and the kind of noise which emanates from it, and become used to it quickly

The two final points which emerge from this case I think are these We are looking for children whose hearing is defective, while they are very young We are hoping to pick them up, if we can, in the first year of life As we do this, the question of finding a proper test procedure is a difficult one, through which I am afraid we are very much fumbling our way, but this case illustrates that it is no use just thinking in terms of some specialized form of audiometry, carried out in the rarified atmosphere of an Audiometric Unit or Department With these young children particularly, there will be a tremendous variation of response to sound, depending upon factors of intelligence and home background We must know about these points before we can really decide whether the child has defective hearing, or whether it is deprived of the normal process of development of understanding of hearing and speech, which begins with the baby in its mother s arms

If we wait until we can make a firm diagnosis of deafness, we are wasting time which is of great value to the child When a normal hearing child may appear to be deaf because it is deprived of a normal hearing environment, we must accept that a partially deaf child will frequently, if not invariably, appear much more severely deaf, and the level of communication may become permanently depressed.

Our best results will follow the institution of remedial training as soon as the defect is suspected. The two main items of training are a well fitting hearing aid and a mother who understands the need to talk to the child and can devote her time and her love to the child.

L. Fisch. I can answer Mr. Crabtree's question as to what would have happened if this child had not been fitted with a hearing aid. The child's language and speech would have developed, as it did, without the aid because of the individual attention and care she subsequently received. The hearing was normal, but there was lack of response to sensory stimulation, as shown by the negative results of the hearing tests at the initial stages of investigation. This type of negative response is frequently observed in babies who have suffered from a serious emotional upset and deprivation.

We have seen many children like this at the Audiology Unit (of the Royal National Throat, Nose and Ear Hospital) including one quite recently. Initially this child did not respond at all to test sounds; she did not use an aid, but when adopted and placed in a happy home she developed normally from the point of view of speech, and finally it was found she had normal hearing.

We have been aware of this type of child for some time, and noticed them particularly among babies who stayed in residential nurseries and suffered from the loss of their mothers. But, although it is known that emotional upset and deprivation can cause this lack of response to various types of sensory stimulation, one must be very careful when examining these babies, for example, when our opinion is asked as part of an examination before adoption. One must remember that these are frequently unwanted children, and there is always a possibility of taking drugs during the early stages of pregnancy for abortion, and this in fact can cause serious deafness.

There is no scientific test of hearing available for these very young children, and the information must be gathered by frequent sessions of observation of the child's responses to various sound stimuli. It is important that the test sounds should represent as wide a range of frequencies as possible. The interpretation of the reactions of the baby to sound stimulation must be made by experienced observers, and these reactions must be carefully recorded. In this way, bits of information can be collected each session, and step by step, and within a reasonable time, a full picture of the hearing ability of the child will emerge. Although the results cannot be expressed numerically, we can always answer, as a result of these observations, the four principal questions, that is:

Is the child profoundly deaf?
Is he severely deaf with a good amount of useful residual hearing?
Has he a moderate hearing loss?
Or is his hearing within limits which could not cause serious delay in language and speech development?

According to the answers to these questions, the necessary remedial steps can be taken.

GENERAL NOTES

THE Editors are anxious to improve the clinical aspect of the Journal and would welcome more reports of interesting cases These clinical records need not be at all elaborate and it would be unnecessary to go minutely into the ætiology or the literature of the subject Senior Registrars have unique opportunities for seeing a large variety of cases, many with unusual features It is in many ways regrettable that these should go unrecorded It is hoped that all those in charge of clinics will help us in this matter and see that such cases are not overlooked

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THE BÁRÁNY JUBILEE MEDAL

IN conjunction with the graduation ceremonies at the Uppsala University, Sweden, the Bárány Jubilee Medal was awarded to Dr Charles Skinner Hallpike, M D , F R C P , F R C S , F R S , Medical Research Council, Otological Research Unit, National Hospital, London

At the ceremony Dr C O Nylén, the successor of Bárány in the chair of Otology at Uppsala University gave an outline of Hallpike's works

The Medical Faculty of Uppsala has awarded this outstanding distinction for 1958 to the eminent and internationally known scientist, Dr C S Hallpike, London By his clinical and experimental investigations he has increased to a high degree our knowledge of the functions of the vestibular system and of optomotorial nystagmus

The Dean of the Medical Faculty then handed the gold medal to Dr Hallpike

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THE INSTITUTE OF LARYNGOLOGY AND OTOLOGY

330 332 GRAY'S INN ROAD, LONDON, W C 1

The Academic Board announces the following

SPECIAL LECTURES AND DEMONSTRATIONS

to be given at the Institute—except where otherwise stated These are open to all members of the Specialty and to postgraduate students No ticket of admission is required

Friday, 24th October, 1958, at 5 30 p m

PROFESSOR C V HARRISON (Postgraduate Medical School of London)—"The pathology of malignant lymphomas "

General Notes

Saturday, 8th November, 1958, at 11.30 a.m.
PROFESSOR LUCIUS RÜEDI (Zurich)—"New developments in the Surgery of Oto-sclerotic Diseases."

Friday, 14th November, 1958, at 4.30 p.m.
MR. T. G. WILSON (Dublin)—ANNUAL ADDRESS—"The modern treatment of deafness in historical perspective".

Saturday, 6th December, 1958, at 11.30 a.m.
D. J. A. SULLIVAN (Toronto)—"Modern temporal bone surgery".

Friday, 12th December, 1958, at 5.30 p.m.
PROFESSOR W. ST. C. SYMMERS (Charing Cross Hospital Medical School)—"Mycotic diseases".

PROVISIONAL LIST OF SHORT COURSES FOR CONSULTANTS AND SENIOR REGISTRARS

3rd and 4th October, 1958—Practical Aural Surgery Class.

17th, 18th and 19th February, 1959—Malignant diseases of the ear, nose and throat.

20th and 21st February, 1959—Pathology of the ear, nose and throat.

10th, 11th and 12th March, 1959—Diagnosis and treatment of the deaf child.

13th and 14th March, 1959—Modern methods of acoustic and vestibular testing.

10th and 11th April, 1959—Practical Aural Surgery Class.

The number of entries is limited in each course.

A detailed syllabus of any of the above can be obtained on application to the Secretary.

SEMON LECTURE

THIS lecture will be delivered by Professor Luzius Rüedi of Zürich in the Barnes Hall of the Royal Society of Medicine on Thursday, 6th November. Subject : Some observations on the Histology and Functions of the Larynx.

DISLOCATION OF THE INCUS DUE TO HEAD INJURY

By Francis Bauer (Liverpool)

In the caption to Fig. 2. on p. 677 of the above paper (*J. Laryng,* August 1958) the preoperative air conduction should have been marked by large black dots, as in the audiogram, ● ● ● and not by fine dots.

The Journal of Laryngology and Otology

(*Founded in* 1887 *by* MORRELL MACKENZIE *and* NORRIS WOLFENDEN)

October 1958

IDIOPATHIC HÆMOTYMPANUM

By J F BIRRELL (Edinburgh)

HÆMOTYMPANUM is still a sufficiently uncommon condition for sporadic cases to be reported, and a search of the literature might appear to give the impression that the discovery of a blue drum is a clinical rarity Credit for the first full description of the condition belongs to Shambaugh who, in 1929, noted two such cases, and illustrated the appearance of the drum with a colour plate Fowler (1939) made a brief reference to a personally observed case, while O'Donnell (1941) described two cases in children, and suggested the name "idiopathic hæmotypanum" for the condition Kler (1948) discussed one case, and Ranger (1949) reviewed the literature, and added observations and investigations in two cases Vior (1951) described a case of blue drum, and Menck-Thygesen (1952) added two cases, one of whom had the condition bilaterally Johnston (1953), in an interesting article, discussed two cases, while further cases have been reported by Simonton (1955), Bourgeois and Natalie (1955), Terkildsen (1956) and Hall (1957), each of whom discussed one case, while Sheehy and McKibben (1956) described three cases These, as far as I am aware, are the only published reports of the condition

Against this total of twenty cases, it may appear to be presumptuous on my part to describe a further eight cases, two of whom showed the condition bilaterally, but the fact that six of them were discovered within a period of twelve months appears either to confirm the view expressed by Menck-Thygesen that the condition may not be as infrequent as has been thought, or to suggest that the ætiological factor may be becoming more common

Sufficient is known about the condition to classify it as a clinical entity, not a clinical rarity. It has a well-defined history, diagnostic clinical features, consistent radiographic pictures and operative findings, and characteristic histological appearances from material in the mastoid cells. It cannot be named with accuracy until the ætiology has been finally established, but it must be fitted into our conception of otomastoiditis.

Case Reports

The first two cases may be dismissed briefly as notes are not now available to me, and as no treatment was carried out.

Case I. A boy, aged 14, was examined on account of deafness and recurring attacks of pain in the right ear over some years, but with no discharge. Examination was negative except that the right drum was of an intensely blue colour, but of normal contour and with a cone of light. Tuning fork tests showed a conductive deafness in this ear, and radiographs revealed extensive pneumatization of both mastoids, the cells in the unaffected side being healthy, but those in the right mastoid being uniformly blurred. This boy was examined in a Cottage Hospital in the Border district of Scotland, and the parents were unwilling to allow surgery.

Case II. A youth, aged 19, was examined at the request of the Ministry of Labour and National Service on account of a history of recurring attacks of pain and deafness in the left ear. The left drum was deep blue in colour with a normal light reflex, and radiographs showed a uniform obscurity of a well-pneumatized mastoid process on this side. He was asked to consult his own doctor with a view to being referred back for surgery, but he has not been seen again by me.

Case III. Nicolas W., was examined on 7.3.56 when he was aged 3, on account of recurring earache in both ears, with discharge from the right ear. At that time the right meatus was filled with pus, a swab of which yielded no growth, and the left drum was normal. He had unhealthy tonsils and adenoids, and these were removed on 4.5.56. He was examined again on 21.5.57 on account of further attacks of pain, confined now to the left ear, but with no discharge. The right drum showed a healed central perforation, while the left was intact, blue in colour, and of normal contour. He was again seen ten days later on account of severe pain in the left ear for one day followed by a sudden blood-stained discharge. On examination a little dried blood-stained material covered the drum, and by 4.6.57 the drum was of a normal colour but was bulging. A week later the drum appeared normal. On 2.7.57 there was a further attack of pain in the left ear, and the drum was again bulging and blue in colour. X-rays showed the right mastoid to be normal, while the cells on the left side were obscured and cellular break-down was reported.

On 10 7 57 the *left mastoid* was opened, and a large coalescence of cells was found near the tip This necrotic area contained thick brown mucoid secretion The mucosa of the antrum was thickened The attic was opened, and the incus appeared to be healthy *Pathological examination* revealed giant cells, cholesterin clefts and hæmosiderin He was discharged from hospital ten days after operation, when the drum was slightly indrawn but of normal colour and it has remained thus The last examination was in May, 1958 when the drum was of normal colour, and there has been neither pain nor deafness in the ear

Case IV Raymond T , was examined on 5 6 56 when he was aged 6, on account of recurring attacks of pain and deafness in both ears, treated by his own doctor on several occasions with oral penicillin and sulphonamides There had been no discharge His drums were indrawn and of normal colour His unhealthy tonsils and adenoids were removed on 27 6 56 He reported again on 11 7 57 on account of further attacks of pain and deafness in both ears, treated by his doctor with chloramphenicol Both drums were intensely blue in colour and were indrawn, and he could only hear a whisper at some 4 in in either ear X-rays showed bilateral sclerotic mastoids On 29 7 57 he came to the operating table, and examination of the drums before operation showed both to be indrawn and of normal colour It was decided however, to proceed with the operation, and the *right mastoid* was opened The cortex was dense, but the mastoid contained an area of necrosis filled with black cellular debris extending from the tip to the antrum and petrosal angle The lateral *sinus plate was involved in this necrotic area, but the wall of the sinus* appeared to be healthy More debris was found in the antrum, and blackish-brown mucoid material exuded through the aditus through which it was aspirated There was a similar infection in the zygomatic cells *Bacteriological examination* showed the fluid to be sterile The *pathological report* showed giant cells cholesterin spaces and hæmosiderin On 2 8 57 the *left mastoid* was opened, and the findings were identical with the other side, even to the lateral sinus being exposed by necrosis The attic was not opened, but aspiration was continued through the aditus until no further black fluid could be obtained Again the fluid was sterile and *histology* showed giant cells and cholesterin spaces By 22 8 57 both drums were normal, and on 3 10 57 he could hear a whisper at 12 ft in either ear

Case V Sydney S , was first examined in 1952 when he was aged 5, on account of deafness, especially in the left ear The left drum was indrawn and of normal colour, and his tonsils and adenoids were removed He was seen again on 13 6 57 after his third attack of pain and deafness in the left ear within a year On this third occasion there had been a sudden discharge of blood and pus The left drum was bulging and of a deep blue colour and he only heard a whisper close to this ear The right drum was normal, and his nose and throat were clear. X-rays of the mastoids showed both processes to be poorly pneumatized with obscurity and cellular break-down in the left side. On 3.7.57 the *left mastoid* was opened and found to contain black necrotic cells and a thick yellow-green discharge. A break-down of the cells in the petrosal angle was cleared out, and the subdural cells were also necrosed. The mucosa of the antrum was thickened; the attic was opened and the incus was seen to be healthy. Glairy blackish-green fluid was aspirated from the floor of the attic external to the incus. The *pathological report* showed cholesterin clefts and giant cells, but no hæmosiderin and no purulent infiltration. By 22.7.57 the drum was of normal colour, and he heard a whisper at 6 ft. in the ear.

Case VI. Patrick O'N., developed a meningitis in 1951 when he was five years old. He was examined on 27.3.52 because of deafness in both ears. The drums were indrawn and of normal colour, and he could only hear a conversational voice at 4 ft. in the right ear and at 2 ft. in the left. It was noted that he had large tonsils and adenoids, but apparently no action was taken. He reported again on 25.11.54 on account of recurring attacks of pain in both ears. Examination again showed indrawn drums and on 10.12.54 the Eustachian tubes were inflated. Both were patent, but the right tube was relatively clearer than the left. Under the same anæsthetic his tonsils and adenoids were removed. On 29.10.57 he was again seen with a history of recurring otitis media and deafness, worse in the right ear than the left. The right drum was indrawn and of normal colour. The left drum was also indrawn, showed a scar anteriorly, some crusting in the attic region, and was blue in colour. Examination of the nose, nasopharynx and throat was negative. X-rays of the sinuses were also negative, but those of the mastoids showed bilateral sclerosis. On 25.11.57 he came to the operating theatre when examination showed the left drum to be indrawn and of normal colour, with an anterior scar and an attic crust. The *left mastoid* was opened and was diploic in type. Black cellular necrosis was found superficially in the angle between the lateral sinus and the middle fossa dura. The antrum appeared to be healthy, but, on opening the attic, the incus was found to be lying loose, and was removed. Both processes were eroded. A large granulation was removed from the attic. The head of the malleus was healthy, and no cholesteatosis was found. The *pathology* showed that the granulation tissue from the attic contained the elements of cholesterin granuloma—cholesterin clefts, giant cells and hæmosiderin. There was no squamous epithelium and no pyogenic infection. As the right ear had been more markedly involved in the previous pain and deafness, the *right mastoid* was opened on 2.12.57, but contained no black cells. Again, the incus was lying loose, and was seen to be eroded after its removal. On 7.1.58 both drums were indrawn and healthy in colour, but he was still deaf On 16 2 58 the Eustachian tubes were inflated, and, again, both were patent

Case VII Graham Y , had his tonsils and adenoids removed at the age of 6 in another hospital in 1956 on account of deafness The hearing improved thereafter, but he was seen on 18 2 58 on account of recurring pain in the left ear, treated by his own doctor with penicillin The nose and throat were clear The right drum was indrawn, and of normal colour. The left drum was also indrawn, but was intensely blue in colour Rinné's test was positive in the right ear, and negative in the left, and Weber's test was lateralized to the left He could hear a whisper easily at 10 ft in the right ear, but only at 1 foot in the left X-ray of the sinuses showed some mucosal thickening in the left antrum, and that of the mastoids revealed marked cellular break-down in the left mastoid, while the right was normal On 7 3 58 the *left mastoid* was operated upon The cortex was dense, but the mastoid contained a cavity filled with thick black mucoid discharge There was extensive break-down of the mastoid cells, and the sinus plate was included in this necrotic area, which extended from the tip to the petrosal angle The antrum and attic contained stringy mucus, chocolate in colour streaked with yellow The mucosa of the antrum and attic was thickened, and the incus was healthy *Bacteriology* showed the fluid to be sterile, and *histology* again revealed the characters of cholesterin granuloma—giant cells, cholesterin spaces and hæmosiderin On 8 4 58 the drum was normal and he could hear a whisper at 20 ft in this ear

Case VIII Barbara D , had her tonsils and adenoids removed in another hospital, when she was aged 5, on account of deafness She was examined on 10 2 58 because of deafness for a year, and attacks of pain in the right ear Her tonsils had been well removed, and her nasal airway was good The left drum was indrawn and blue in colour The right drum was acutely indrawn and intensely blue She could only hear a whisper close up in either ear X-ray of the mastoids showed both to be poorly cellular, with no break-down On 21 3 58 examination before operation showed both drums to be indrawn and of normal colour The *right mastoid* was opened and found to be diploic in type with a few black cells near the tip There was no break-down of the intercellular walls, but the mucosa of the antrum was thickened and stringy mucus was present in the cells throughout The attic was opened and the incus was healthy *Pathological investigation* failed to reveal any of the features of cholesterin granuloma In view of this, the other mastoid was not operated upon

These case reports, and the operative findings and pathological appearances in Cases III–VIII, bear such a striking resemblance to each other, and to the other published reports that one can, with some certainty, dogmatize on the features of the average case One may assume that Case VIII showed the condition in the earliest phase with only a few black cells and negative pathology, while Case VI, in whom the incus was eroded and a large granulation was present in the left attic, represents the most advanced pathology.

History

A history of *pain or discomfort* in the affected ear has been a feature common to all cases. Earache, of course, is a frequent symptom in childhood, occurring bilaterally or in either ear at different times, but recurring attacks of pain confined to one ear are much less common and require particular investigation. Two points emerge from the histories detailed above. In the first place it is noticeable that in many of the children there was a history of bilateral otitis media, and that, after removal of tonsils and adenoids, the pain in one ear disappeared, and at a later X-ray examination this mastoid appeared healthy. On the other hand, the operation had no effect on the recurring attacks of earache in the ear in which the drum eventually became blue. This is in contradistinction to O'Donnell's experience in which removal of tonsils and adenoids cured the condition of blue drum.

The second point emerging from the history is that these attacks of earache have been treated by the family doctor with oral antibiotics. Efforts have failed to ascertain the exact state of the drum during these exacerbations, the exact antibiotic used, and the exact dosage of that antibiotic. One feels that the antibiotic currently popular at the time of examination has been the one used, irrespective of the sensitivity of the causal organisms. One feels, too, that the dosage may possibly have been inadequate, and that no very definite criteria of cure may have been observed. Masking may thus follow, and the symptoms may well recur, due to a focus of infection lying within the mastoid cells, the middle ear or the Eustachian tube.

Aural Discharge. This is not an invariable feature, and was only noted in two of my cases. In each it was described by the mother as being sudden, short-lived and blood-stained. The fluid is presumably the same as that obtained at paracentesis or aspiration of the middle-ear cavity, and this has been shown by Ranger to be essentially altered blood with a positive Van den Bergh reaction. Culture of such fluid is usually sterile, but Shambaugh found *Staphylococcus albus* in his cases. My two cases in whom there was a history of spontaneous discharge were not seen until the discharge had ceased, and no swab could be taken.

Deafness is a complaint common to all the histories, and it is noteworthy that this persisted in some measure between the attacks of pain. It varies from a muffled feeling reported in adults to a deafness complained of by the parents of the children. My own tests suggested that it was conductive in type, and this view is supported by O'Donnell, and Sheehy and McKibben Ranger's audiograms show a fairly flat curve, as do those of Kler, Johnston and Simonton Menck-Thygesen's audiograms, on the other hand, suggest a mixed type of deafness with a greater loss for the higher notes Improvement in the audiograms is slight after myringotomy and more marked after mastoid operations

Examination

As a rule, examination has been negative apart from the blue drum in the affected ear and the radiographic evidence of mastoid involvement on that side In some cases, however, the history or clinical features in the nose or nasopharynx have led me to have the sinuses X rayed, but the films did not show any consistently present abnormality

The blue drum is characteristic of the condition, and is typically of a dark colour Sheehy and McKibben have emphasized the distinction between this and the gun metal blue of serous otitis media Shambaugh remarked on the fact that the blue colour was confined to the pars tensa, while the pars flaccida was of normal colour, and this is not to be wondered at in view of the cause of the discoloration All varieties of contour of the drum have been observed in association with this blueness, indrawing, normality and bulging The jugular vein on the same side should be compressed as a diagnostic measure, because if the colour were due to the jugular bulb an alteration in the appearance of the drum should be observed

I examined three of my cases on the operating table immediately before operation after they had spent 24 hours in bed, and in none was the blue colour evident Yet the characteristic black cells were present in the mastoids of all three cases at operation This raises the question, which will be discussed later, as to whether the blue appearance is transitory or not

The Eustachian tube It is not surprising that the Eustachian tube should have been blamed for the deafness in these cases, and, indeed, for the source of the blood, even though many authors have commented on the ease with which tubal inflation could be carried out Shambaugh and Fowler both declared that the tube was patent, Kler stated that the Eustachian tube could be inflated easily but noted that this produced no improvement in the deafness Menck-Thygesen reported that there was no tubal occlusion at the outset of one of his cases, but that this developed later, and was therefore not the cause of the deafness Johnston also noted patent Eustachian tubes, while in two of Sheehy and McKibben s three cases the tubes were clear The tube was blocked in the third case

Blood investigations have been carried out by a number of authors and have been largely negative The presence of cholesterol clefts in the histological sections has led to the serum cholesterol being investigated

The difficulty of this is that there is a wide normal variation depending, among other things, on the technique employed, and differing in different laboratories. Ranger pointed out this normal variation but concluded that his patients showed no disorder of the blood which would give an increased tendency to bleed. Johnston found a normal blood picture and blood chemistry apart from a high prothrombin concentration of 70 per cent. (normal 30 per cent.). His patient had a serum cholesterol of 215 mg. per 100 ml., the normal being 110–390 mg. per 100 ml. Kler's patient had normal bleeding and clotting times, normal prothrombin and platelet count, and the tourniquet test did not reveal any fragility of the capillaries. Hall, who thought at the operation that the affected mastoid area was unduly vascular, could find no corroboration of this histologically or angiographically.

Mastoid radiography. It has been the universal experience of those who have had the mastoid processes X-rayed that the films have shown cloudiness on the affected side. Most of the films showed cellular processes, but in some pneumatization was not prominent. In the cellular mastoids, the cells were homogeneously opaque, and not infrequently there was well-marked evidence of destruction of the intercellular walls.

Treatment

Removal of Tonsils and Adenoids. O'Donnell stated that his two cases were cured of their deafness, and the blue colour of their drums gradually disappeared, following removal of the tonsils and adenoids. No other author has had a similar experience. It will be seen that six of my cases (Cases III–VIII) had had their tonsils and adenoids removed for a history of bilateral earache and deafness, and that at the time of that examination the drums were not blue. The operation relieved the condition in one ear, as one would expect, but the recurring attacks of earache and deafness persisted in the other ear, and this drum subsequently became blue. From this it would appear that the removal of tonsils and adenoids can have had no bearing on the progress of the condition, assuming that it was developing at that time, and that the source of the blue drum does not lie in infection of the lymphoid tissue of Waldeyer's ring.

Inflation of the Eustachian tubes. Inflation of the Eustachian tubes alone has no effect on this condition. As has already been stated, many of the authors mentioned that the tubes were patent, and Kler has asserted that tubal inflation does not improve the hearing. Again, O'Donnell is in the minority as he states that there was some obstruction to inflation by catheter. On inflation he reported that râles were heard, and afterwards the hearing for a whisper improved from 6 inches to 10 feet. The colour of the drum changed to a dull grey with brownish patches.

Myringotomy. This was the sole method of treatment until 1949. The drum was incised and thick chocolate coloured fluid was aspirated.

Tubal inflation assisted in expelling the thick secretion from the middle-ear cavity. Analysis of this fluid has shown it to be altered blood, and most authorities have found it to be sterile. Shambaugh reported *S. albus* from the secretion. The result of myringotomy was a temporary relief from the symptoms and a slight improvement in hearing. The drum either remained blue (Hall) or quickly returned to its blue colour, and the process had to be repeated, either by myringotomy or by aspiration through the drum by means of a needle. Menck-Thygesen noted that the drum became anæsthetic, and aspiration could be performed without any local analgesia. Hall on the other hand states that the drum was so tender that pain was experienced on touching it with cotton wool, but this he ascribed to neurosis on the part of the patient.

Mastoid operation. The failure to effect a permanent cure by these methods led inevitably to an attempt at mastoid surgery, and Ranger, who was the first to describe this, gives a clear account of the removal of black debris from the mastoid cells, leaving an apparently healthy cell lining. He performed the cortical (Schwartze) operation and found that one of his cases had an extensively pneumatized mastoid, and the other a smaller acellular process. Since that date (1949) Menck-Thygesen, Bourgeois and Natalie, Johnston, Terkildsen, Sheehy and McKibben, and Hall have all used the cortical operation with success, while Simonton and Sheehy and McKibben have had to proceed to the radical operation, in one case each, in order to obtain a cure. My own experience has been confined to the cortical mastoid operation in Cases III–VIII without any previous attempt at myringotomy. This treatment has been based on the clinical findings of a blue drum together with radiological evidence of mastoid involvement. I can see no place for myringotomy in the treatment of the condition if, as seems certain, the source of the blood is the mastoid process. The extensive involvement of the mastoid process, and the cellular necrosis found at operation makes it obvious that nothing short of mastoid surgery can cure the condition. The results obtained by the operation appear to justify this view. The hearing improves, the pain disappears and the drum returns to its normal colour and remains thus.

Bacteriology

Bacteriological examination of the secretion in the mastoid cells shows it to be a sterile fluid. In the main the cases have been operated upon in the quiescent phase, and it may be argued that this sterility has been due to the antibiotic drugs used by the general practitioners. Case VII was operated upon immediately after an exacerbation of pain, but the secretion was sterile.

Pathology

Histological investigations of the mastoid cells and their black cellular content have consistently shown the presence of giant cells and cholesterol spaces. Less consistently, hæmosiderin has been found. Such a pathological picture is now known as cholesterin granuloma, and is not necessarily confined to patients with blue drums. I have previously described a number of cases exhibiting true cholesteatosis in the attic and cholesterin granuloma in the subcortical cells (1956).

Discussion

Johnston and Vior have discussed the possible causes of a blue appearance of the tympanic membrane, and the following have been mentioned (1) a fracture of the base of the skull, (2) the mechanism of the decompression chamber, (3) varices of the middle ear, (4) hæmangioma of the middle ear, (5) protrusion of the jugular bulb and (6) hæmophilia. There is no doubt that any of these could result in a blue drum, and there may be many more similar conditions, but it is quite obvious from the eight cases which I have recorded, and from the majority of the cases reported in the literature, that the condition described has a separate ætiology, and can be readily differentiated from the above by the history, blood examination and radiography.

It is generally agreed that the colour of the drum is due to the presence within the middle ear of a thick fluid which is essentially altered blood. The source of this blood must be the Eustachian tube, the middle-ear cavity or the mastoid cells. Earlier opinion favoured the tube or the middle ear, and it was only after similar fluid was found in the mastoid cells that it came to be recognized that these cells were the most probable site of origin. Hæmorrhage in the mastoid, as Ranger pointed out, would produce sufficient fluid so that it would be difficult to clear it by aspiration through the drum, whereas hæmorrhage arising in the tube or middle ear might easily be so cleared. Again, hæmorrhage in the middle ear or tube would not be cured by mastoid operation. The destruction of the intercellular walls, and the extent of involvement of the mastoid cells is such that it appears obvious that the mastoid is primary site of trouble.

One must therefore discount earlier theories, based on the assumption that the source of the blood is in the tube or middle ear. Kler's view that there is a congenital anomaly of the veins draining the tympanic membrane to permit an escape of a minimal amount of blood into the membrane and middle-ear cavity, and Johnston's theory of an area of varicosity or even hæmangioma in the middle-ear cleft which fails to undergo complete tissue repair, but which repeatedly breaks down to permit of the escape of fresh blood, may account for the occasional blue drum, but cannot be accepted as the common cause. Ranger examined O'Donnell's theory of a *hyperæmia ex vacuo* and rejected it on three counts (*i*) Eustachian obstruction is common but hæmotympanum is rare, (*ii*) Eustachian obstruction is not found in all cases of hæmotympanum and (*iii*) very few cases of *hyperæmia ex vacuo* have been found in acute otitic barotrauma occurring in aviators or experimentally produced in cats

Menck-Thygesen places the origin of the bleeding in the mastoid cells, as do Sheehy and McKibben who added that surgery had so far failed to uncover the source of the bleeding Hall thought that the affected areas in the mastoid were unduly vascular, but was unable to confirm this histologically

I have been interested to find that examination of three of my patients before operation failed to show the blue drum which had previously been apparent in the out-patient department, and, that in spite of this, typical appearances were found in the mastoid process and typical sections were obtained histologically It is evident from this observation that the fluid is not sealed within the middle ear, but is able to escape, either along the relatively wide Eustachian tube of the child or backwards through the aditus into the mastoid antrum

Continuing with this line of thought, it might therefore be possible for the condition of cholesterin granuloma to exist in the mastoid of an ear in which the drum was consistently of a normal colour on examination If this were so, it would be natural to expect that, if a number of mastoids were to be opened in children with a similar history and similar radiological appearances to those described but with normally coloured tympanic membranes, one should be able to demonstrate black cells and cholesterin granuloma in a significant percentage of them Ranger, indeed, quoted such a case of secretory otitis media in which cholesterol clefts were found histologically, but in an amount less than in cases of hæmotympanum

During the past two years I have operated upon twenty children with histories of recurring attacks of pain and deafness in one ear, with radiological evidence of opacity of the mastoid cells and break-down of the intercellular walls in the same side, but whose drums, on repeated examinations have been entirely devoid of any blue discoloration Space will not permit of a detailed description of each case and, indeed, such descriptions would be largely repetitive Two may be cited as typical examples

Douglas S, was seen on 27 4 54, aged 5, with a history of recurring attacks of bilateral otitis media and deafness treated by his own doctor with oral penicillin His tonsils were small and slightly congested, and he had enlarged cervical glands His nose was clear Both drums were indrawn, but of normal colour He could only hear a whisper at 1 foot in either ear On 10 5 54 the Eustachian tubes were inflated, and were found to be patent, and his adenoids were removed Six months later he was seen on account of further bilateral otitis media and deafness The right drum was acutely indrawn and congested and the left drum was normal On 18 2 55 his tubes were again inflated with ease, and his tonsils and some adenoid tissue were removed. A year later he again reported on account of several further attacks of right-sided otitis media. Again, the right drum was acutely indrawn and congested and an X-ray of the mastoids showed diploic processes with cellular break-down in the right side. On 30.3.56 the *right mastoid* was opened and the cells were found to contain clear, faintly yellow, tenacious mucus. There was an area of cellular necrosis from the tip to the antrum. *Bacteriology* showed that the fluid was sterile. *Pathological investigation* revealed œdematous mucosa infiltrated by lymphocytes and plasma cells, but with scanty polymorphs. No features of cholesterin granuloma were present. Eighteen months later, on 28.11.57, he had had no further trouble with the right ear, the right drum was only slightly indrawn, and he heard a whisper at 16 feet in this ear.

Sadie G., had her tonsils and adenoids removed elsewhere in 1952, when she was aged 4. She was examined on 21.10.54 with a history of colds, catarrh, earache and deafness. Her throat was clear but her nose contained mucopus. Both drums were indrawn and healthy in colour. X-ray showed both antra to be opaque. On 1.11.54 a double proof puncture yielded no pus, and some adenoid remains were removed. On 5.5.55 she reported back with a similar history, and the clinical examination was unchanged. X-ray on this occasion showed an opaque left antrum, and this was punctured on 20.5.55, again yielding no pus. She was examined on 30.1.56 on account of deafness and recurring pain in the left ear. The left drum was congested and indrawn and an X-ray showed blurring of the cells of the left mastoid. On 6.5.56 the *left mastoid* was opened, and the cells were found to be filled with thick, sticky mucopus, which was sterile. Pale, adherent granulations were present in a necrotic area extending from the tip to the antrum. The antral mucosa was thickened, and, on opening the attic, a healthy incus was found embedded in thickened mucous membrane. *Pathological* features were identical with the former case. On 11.7.57 her mother reported that Sadie was in much better health, her school work had improved, and her hearing had greatly increased. Both drums were slightly indrawn and of a normal colour.

In none of this type of case has a single black cell been found in the mastoid process. In none has there been any suggestion of cholesterin granuloma in the histological sections. All have shown evidence of infiltration of the mucosa by lymphocytes, polymorphs and plasma cells, while in some cases fibrosis has been noted. The picture is that of a chronic non-specific inflammation.

Thus, in spite of the similarity of the history, in spite of the similarity of radiological appearances, in spite of the mastoid necrosis involving the same groups of mastoid cells, the two conditions of hæmotympanum and secretory otitis media are different. I am persuaded, however, that the ætiology of the two conditions must be closely allied, because the only difference is the presence of blood in the one, and not in the other.

Robison (1942) investigated catarrhal otitis media and mastoiditis with effusion, and divided it into two types. The first is due to some demonstrable lesion in the upper respiratory tract, correction of which brings relief of the aural symptoms. The second type does not appear to be associated with upper respiratory tract infection. The tympanum is entirely or partly filled with fluid which may be seen through a retracted membrane. According to Robison this membrane has a yellowish or bluish colour depending on the colour of the fluid in the tympanum. The Eustachian tube is usually partly closed on inflation. Investigation of the total protein in the fluid aspirated from the middle ear convinced him that it was a transudate and not an exudate. He advised mastoid surgery, if other treatment fails, on account of an impending loss of hearing, and with this opinion I am in entire agreement.

Cody (1941) described four cases of catarrhal oto-mastoiditis very similar as to operative appearances and histological findings to the twenty on whom I have operated. He found that their Eustachian tubes were patent, and concluded that "it was obvious that the middle ear and Eustachian tube were the passageway, not the place of origin, of the mucous secretion". My experience in these secretory cases, which come to mastoid operation, leads me to support this opinion. The source of the fluid lies in the mastoid cells, and not the middle ear or tube.

Secretory otitis media, like hæmotympanum, has become much more common in the past fifteen years, and Eagle (1946), commenting on a series of 172 cases seen in the previous four years writes, "We have no definite causative factor for the increased frequency of the disease . . . It is possible that we are dealing with a virus disease; since the disease is almost in epidemic form, there is reasonable ground for this suspicion. We have been unable to correlate the use of sulphonamides in recent years with the disease . . . We do not admit that the disease is necessarily associated with upper respiratory disease. We are reluctant to admit that the disease had been prevalent prior to 1941, and that our eyes are now seeing pathology which they had previously failed to see."

Bateman (1957), reporting 67 cases of secretory otitis media, found that in extreme cases the drum appeared blue (presumably the gun-metal blue noted by Sheehy and McKibben). He found that the fluid from the middle ear was sterile, and was of the opinion that it was a secretion and not a transudate, an opinion which conflicts with that of Robison. He also noted that puncture-aspiration through the drum could be performed with little or no surface anæsthesia.

In secretory otitis media, as in hæmotympanum, the source of the fluid appears to be the mastoid cells. Why does it not drain away? If the Eustachian tubes were blocked in every case it would be easy to understand this, but Cody asserted that they were patent, Robison stated that they were partly blocked, and Bateman found that, while his patients could not clear their own tubes, they could be inflated by Politzerization. The majority of opinion in hæmotympanum, as we have seen, is that the tubes are patent.

Is it that the fluid is produced in too great a quantity? If one performs a cortical mastoid on a patient with an intact drum, and closes the post-auricular incision completely, the blood within the mastoid cavity will clot, but some of it must have entered the middle-ear cavity and Eustachian tube. Therefore, the drum should appear blue at the end of the operation, and possibly for a day or two thereafter. Yet it is uncommon to find a blue drum when the mastoid is dressed a week after operation, and such blueness quickly vanishes. I have invariably closed completely the incisions of those mastoid operations performed for hæmotympanum and catarrhal otitis media, and I have never seen a drum appearing blue on the patient's discharge from hospital 10–14 days later. One must, therefore, assume either that the operation has allowed the tube to open, or that the Eustachian tube, whether partly blocked or not, can drain from the middle ear and mastoid cavities the fairly considerable volume of blood produced at operation.

The one noteworthy feature of the fluid in the mastoid cells at operation for secretory otitis media or hæmotympanum is its viscosity, and it may be that this quality hampers normal drainage through the Eustachian tube in these cases. Once removed at mastoid operation, it must not re-form, or the condition would surely recur.

It seems reasonable to suppose that something might be learned about these conditions from microsurgery if the drum is reflected as for a mobilization of the stapes, but Sheehy and McKibben's third case, in whom the drum remained blue after operation, was so examined and the middle-ear cavity and tubal entrance were found to be healthy.

Ætiology of hæmotympanum still remains uncertain in the typical case in which angioma, fractured skull, jugular bulb abnormality, or blood dyscrasia can be excluded. Should Eagle's suggestion that a virus may be responsible for secretory oto-mastoiditis be correct, it is possible that a virus which stimulates a hæmorrhagic response in the tissues may account for hæmotympanum. The virus of influenza springs to mind as a possibility, as it is associated with hæmorrhagic bullae on the drum, with epistaxis, and even, in the recent epidemic, with spontaneous septal hæmatoma. Such a virus infection would not be fully controlled by penicillin, although this antibiotic would prevent invasion by secondary pathogens, which might account for the sterility of the fluid. Repeated exacerbations from upper respiratory tract infections might be responsible for the mastoid necrosis which develops.

It is easier to refute theories than to propose new ones, but I think that one may discount the view that the source of the blood is either the Eustachian tube or middle-ear cavity. One may also, on the evidence of published literature, discard the idea that a blocked Eustachian tube is responsible for the condition. The mechanism is not that of acoustic barotrauma. Nor, as far as can be ascertained, can an abnormal serum cholesterol be demonstrated. It is a confession of ignorance that the term "idiopathic" must still be applied to the condition.

Prognosis

One is left to contemplate the prognosis had mastoid surgery not been undertaken. Would the necrotic area in the mastoid process heal, or would the condition progress slowly? In 1956 I reported a series of 21 children in whom cholesterin granuloma (then mistakenly called black cellular cholesteatosis) was present in the mastoid cells. Two of these cases showed no perforation: four had developed a postero-marginal perforation with granulations: fourteen showed a perforation of the membrana flaccida with the formation of granulations or a polypus: while one patient had a large polypus, the exact site of origin of which was not determined. Of these 21 children, frank cholesteatosis, with typical histology, was present in ten. One of these children was the older sister of Case III in the present series.

The most advanced pathology of the cases here reported was found in Case VI, which showed a blue drum with an anterior scar and a crust over Shrapnell's membrane. The incus was eroded and was lying loose, and from the attic was removed a large granulation, section of which revealed the typical features of cholesterin granuloma. It appears to be within the bounds of possibility that, if this mastoid had not been operated upon and the granulation removed, perforation of the membrana flaccida and eventual immigration of squamous epithelium with the production of cholesteatosis might have taken place.

It is too great a coincidence to ignore that black cells and cholesterin granuloma may be present with an intact blue drum and also with perforation and cholesteatosis. I am convinced that mastoid operation on these children not only cures the condition and improves the hearing, but may prevent perforation in Shrapnell's membrane, or near the drum margin, with all its consequences.

Summary

The condition of hæmotympanum may not be as uncommon as would appear from a review of the literature. Eight new cases are reported, six of which have been seen in children in the past year. The similarity of their histories, clinical features, radiological findings, operative appearances, and pathological reports classifies them as forming a clinical entity.

All have been cured by cortical operation in preference to myringotomy.

Many conditions may cause a drum to appear blue, but not all are associated with radiological mastoid opacity, and cellular breakdown with black cells in the mastoid process, and with histological evidence of cholesterin granuloma.

The various ætiological theories are examined. The source of the blood is the mastoid process. Comparison with secretory catarrhal oto-mastoiditis leads one to suspect a common ætiology, and, if a virus infection is a possible cause of this condition, a virus resulting in a hæmorrhagic response may account for hæmotympanum.

It is suggested that, if the condition is not treated surgically, cholesterin granuloma may form in granulation tissue in the attic, and lead to perforation, immigration of squamous epithelium, and attic cholesteatosis.

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SOME POST-OPERATIVE HAZARDS IN TRACHEOSTOMY ON INFANTS

By PHILIP READING (London)

Tracheostomy on infants is attended by considerable risk during the immediate post-operative period, de cannulation provides a further, if remote, hazard Of necessity performed by any surgeon who happens to be at hand, the operation is perhaps not always conducted with that nice attention to detail which is so carefully enjoined in laryngological text books Outside the special departments of large hospitals the operation is relatively uncommon, so that the nurse placed in charge of the tracheostomized infant may never have seen a similar case—and no amount of theoretical instruction can compensate for lack of practical experience—and yet it is common enough to provide a recurring source of anxiety Remembering the infant's inability to tolerate even slight degrees of anoxia and the narrowness of its air passages, one is not surprised that serious complications occur more frequently in early than in later life

In the following case history most of the graver complications arose, a study of certain features (italicized) in the tragic post-operative period may throw light on the causation of these complications and help to avoid them in future

M B , aged six months, was under the care of the Pediatric Department for investigation of screaming attacks which had been present since birth, January, 1955 After a normal pregnancy the child presented as a breech, having to be delivered by the application of forceps to the after-coming head, he was deeply shocked at birth but improved slowly after laryngeal intubation and aspiration and the administration of intra-gastric oxygen For the rest of his life he was subject to intermittent, generalized muscular twitching

On July 13th, 1955 he was admitted, his intense screaming attacks occasioned *spasms of stridor, during which he was noticed to lie with his head and neck markedly extended,* almost in the attitude of opisthotonos Developing a feverish illness which was diagnosed tentatively as pertussis, he was sent home for a few weeks

On September 14th he was re admitted for exacerbation of his inspiratory stridor, tracheostomy over an indwelling bronchoscope was performed a few days later, with immediate but short-lived improvement in the breathing. *The day after operation respiration again became stridulous and laboured: the tracheostomy tube had slipped out of the tracheal lumen and was lying in the cellular tissue in front of the trachea.* Before this accident was recognized, *surgical emphysema had spread over the root of the neck and over the chest wall, and bilateral pneumothorax had developed* (Fig. 1). A new tube was introduced into the tracheostomy with some difficulty, 40 c.c. of air were extracted from the right pleural sac and bronchoscopic aspiration was performed. By the end of the week, the right lung had completely re-expanded, but there was still a small pneumothorax on the left.

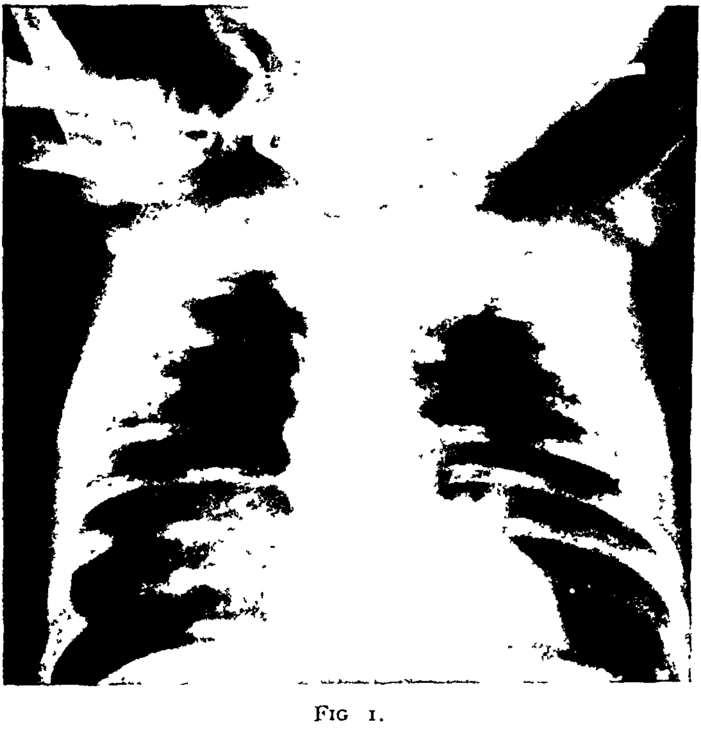

FIG 1.

Tension pneumothorax, complete on right, partial on left, after tracheostomy.

As soon as the chest had returned to normal, repeated attempts were made to de-cannulate the child, but in vain. On every occasion that the tube was removed, *the airway through the glottis proved insufficient to permit normal breathing.* Direct laryngoscopy showed the vocal cords to be normal in appearance and movement, but in the *sub-glottic area the lumen of the trachea was narrowed* from before backwards. Two weeks before death in November, 1955 an inflammatory mass developed over the anterior surface of the trachea in the suprasternal area. During the six weeks of his illness, *the child lay with the head and neck markedly extended* most of the time

Post-mortem examination showed the cause of death to be bronchopneumonia Quoting from the pathologist's report "On the anterior wall of the trachea, its upper extremity lying 14 mm below the lower border of the tracheostomy opening, was a circular ulcer, 12 mm in diameter, with a yellow, sloughing base and slightly raised edge It was non-specific in type and presumably traumatic in origin Further dissection of the upper respiratory tract after fixation revealed the following changes (Fig 2)

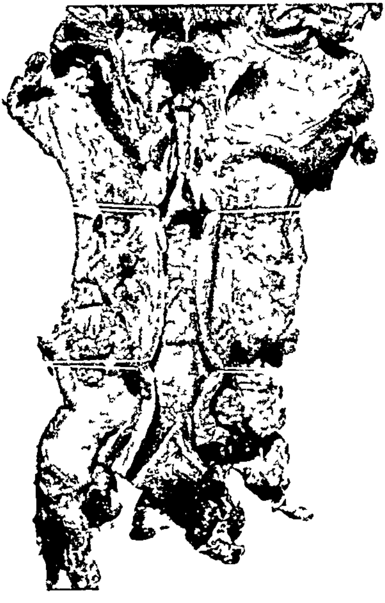

FIG 2

Larynx and trachea opened from behind showing backward displacement of tracheal wall above the tracheostome and an extensive ulcer below

(*a*) The tracheal ulcer had completely eroded through the anterior wall of the trachea over several rings On the outer aspect of the wall superficial to the bed of the ulcer was a dense mass of collagen 8 mm. thick.

(*b*) Immediately above the tracheostomy there was backward displacement of the anterior tracheal wall resulting in narrowing of the natural airway. No structural derangement could be found in the larynx itself."

Examination of the brain showed pseudo-calcification of the lateral nuclei with severe gliosis and collection of foamy macrophages around the capillaries. The examining pathologist concluded that, while the manner in which these brain lesions were produced was not certain, they had probably resulted from multiple foci of ischæmia due to acute brain swelling following birth trauma, the pattern of the lesions being unlike that resulting from generalized anoxia of the brain.

Thus we had been treating an infant who had suffered birth injuries to the central nervous system, a result of difficult and protracted labour. There had been a laryngeal obstruction due probably to derangement of the normal neuro-muscular control; the tracheostomy designed to relieve the obstruction had met with disasters in the post-operative period, and these, not in themselves the cause of death, had yet contributed materially to morbidity and indirectly to the child's death. A study of certain features in this case-history may be of value and interest.

"The day after operation the respirations again became stridulous and laboured."

The care of the tracheostomized infant is often shared between the pædiatric physician and the surgeon, a dangerous division of responsibility, unless both practitioners appreciate the over-riding duty of the surgeon to ensure that at all times the child receives an adequate amount of air through the tube.

When in the course of the illness laryngeal obstruction first became evident, it was manifested by rapid, laboured breathing, by inspiratory stridor and by inspiratory recession of the root of the neck and intercostal spaces, signs calling urgently for relief. After the tracheostomy there may be a falling off in alertness of medical and nursing attendants. The operation has been done—the re-appearance of rapid, laboured respirations, of cyanosis and even of recession of the chest wall is consequently dismissed as being due to broncho-pneumonia. These signs should immediately be regarded in the first place as being due to blockage or slipping of the tube. In neglect of the apparently trivial minutiae of after-care do disasters such as mediastinal emphysema and pneumothorax find their origin. "Paradoxically the patient is in greater danger of dying from asphyxia after tracheostomy than he was before it" (Reading, 1949).

Analysing the indications for tracheostomy in 150 consecutive cases (Fitz-Hugh and Morgan, 1956), distinguish sharply between the symptoms of a fixed obstruction in the upper airway, giving rise to rapid and obvious anoxia, and a fluid obstruction in the lower air passages causing a slow, obscure anoxia The signs of this condition are even more often overlooked after operation, and they demand active surgical measures in the shape of bronchoscopic aspiration as well as the antibiotic therapy which the physician has probably already administered in vain

In estimating the functional lumen of the larynx, Gabriel Tucker (1932) has shown that sub glottic measurements are of greater significance than those of the larynx itself He found the antero posterior diameter of the infantile larynx at the level of the glottis to vary between 7 to 9 mm , the diameter of the sub-glottic space, however, was between 6 and 8 mm He stated that a sub glottic diameter of 5 mm was too small, and one of 4 mm represented an actual stenosis In addition the sub-glottic space, being completely surrounded by the rigid ring of the cricoid, can afford no room for soft tissue expansion except at the expense of the airway

An infant, wearing a tracheostomy tube of 18 French gauge is breathing through a tube of 6 mm diameter, if a uniform coating of exudate of only 1 mm in thickness should cover the walls of the tube, the available airway is only 4 mm in diameter, which Tucker regards as being equivalent to an actual stenosis

"The tracheostomy tube had slipped out of the tracheal lumen, and was lying in the cellular tissue in front of the trachea"

This slipping of the cannula is a fruitful source of serious complications If there is stertor, or inspiratory stridor, and if the hand placed an inch or so from the mouth of the tube does not feel the expired air as a definite jet of gas, the patency and the correct position of the tube must be suspect Superficially all seems to be well—the shield of the tracheostomy cannula lies normally against the neck, air is heard to escape, and there is a tendency to assume that the air is issuing from the lumen of the tube If the tube is in fact displaced, the soft tissues of the neck and the edges of the tracheal incision are sucked together on inspiration, air can enter the trachea only through the diseased, narrow larynx, and it shows a greater tendency to be drawn into the soft tissues of the neck On expiration some air passes through the vocal cords, and some blows a path for itself through the layers of the wound and so reaches the exterior around, but not through, the tube It is this escape of air which may delude the unwary observer into thinking that normal respiration is being carried on through the tube

Slipping of the tracheostomy cannula has been attributed to post-operative swelling of the soft tissues, the œdema forcing the shield of the tube in one direction and the trachea in the other But the position of the neck has a large part to play in determining the accident

Extension of the neck not only makes the trachea more superficial and accessible to the surgeon (Fig 3), it also draws several additional tracheal rings out of the chest (Fig 4) If the tapes securing the tracheo-

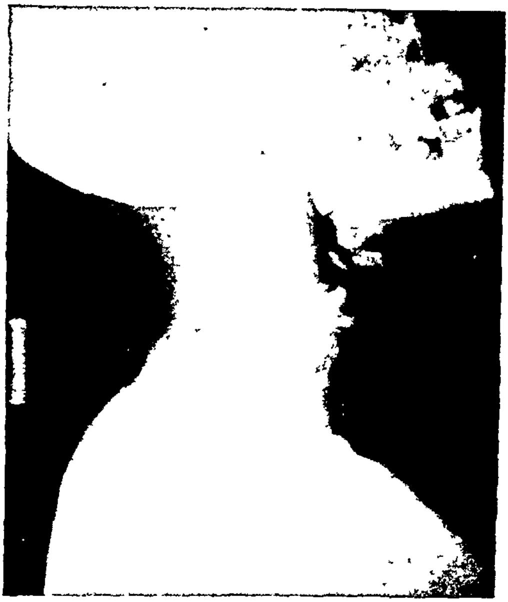

FIG. 3
Head erect, five rings of trachea can be counted.

FIG. 4.
Head and neck fully extended, two further rings are drawn out of the chest.

stomy tube are tied while the neck is still extended, two things may happen when the neck flexes into a more natural and comfortable position. Firstly, the trachea will sink together with its tracheostome. The tube being anchored to the neck (Fig. 5), its lower end may be withdrawn from the lumen of the trachea. If the neck has been strongly extended, and if the incision into the trachea has been made at too low a level, the opening may sink into the chest, giving rise to mediastinal emphysema, as pointed out by Holinger *et al.* (1955).

FIG 5

Head and neck flexed, only one tracheal ring can be seen, and the trachea has receded from the surface

Secondly, when the neck is flexed, the tapes, tied when the neck was extended, will become loose, because the diameter of the neck is reduced. It is suggested that loose tying of the tapes with the neck in extension is a common cause of slipping of the tube.

"*Surgical emphysema had spread over the root of the neck and over the chest wall, and bilateral pneumothorax had developed.*"

Forbes, Salmon and Herweg (1947) reviewing the clinical course of 120 tracheostomies in young children, found that 25 per cent. developed mediastinal emphysema and 10 per cent. pneumothorax, detected clinically. The onset of these symptoms is of most serious significance. Neffson (1949) states that the death rate in unilateral pneumothorax after tracheostomy is 25 per cent. and in bilateral 89 per cent.

Samuel Wilks, physician to Guy's Hospital, wrote in 1860: "We believe there is another source of danger in tracheotomy, but, as we have never seen this alluded to by authors, we would speak of it with some hesitation from our small data. We refer to pneumothorax arising from the escape of air from the mediastinum, having been drawn into the thorax from the neck." He recorded three deaths after tracheostomy; at autopsy all had mediastinal emphysema and collapsed lungs.

Champneys in 1882 carried out experiments on the bodies of still-born children. After performing a tracheostomy he inserted a cannula into the trachea, and then did strenuous artificial respiration. Of 21 bodies so experimented on, 7 showed mediastinal emphysema, and of these 5 had collapsed lungs. From this work, and from post-mortem studies on a series of patients who had died after tracheostomy, he concluded that the emphysema was due to inspiration of air direct through the cervical incision, the complication being favoured by the use of a low cervical incision and by wide and protracted dissections of fascial planes in the presence of violent inspiratory movements.

The condition seems to have excited very little attention until 1940. For instance, Figi (1934) described in detail 200 cases of tracheostomy, but did not mention emphysema and pneumothorax as a complication. From an analysis of the 18 deaths in his series, he came to the conclusion that broncho-pneumonia was the usual cause of death. His post-mortem findings might possibly be erroneous; the so-called broncho-pneumonia might really have been collapse of the lung due to the pressure of an undetected pneumothorax.

Barrie (1940) pointed out that, when the thoracic cage is opened at autopsy in the ordinary way, the presence of a pneumothorax might easily escape detection. He demonstrated in four patients who had died from tension pneumothorax after thyroidectomy, that the air had leaked into the pleural sac through the emphysematous mediastinal pleura.

Goldberg, Mitchell and Angrist (1942) reported on the clinical aspects of tracheostomy-induced pneumothorax, on a material of six cases. They adduced experimental proof that air is actually sucked into the mediastinum from the cervical incision. Air injected into the necks of corpses tracked down into the superior mediastinum. A sudden drop in the pressure needed to inject the air indicated that a bleb had burst into the pleural sac. They found, in agreement with Barrie, that this rupture occurred most commonly through the mediastinal pleura near the thymus. Chance movements of the body sufficed to block this opening, whereupon continued injection of air into the neck brought about another leak into one or both pleural sacs.

Their suggestion that the inspired air is trapped below the thoracic inlet by expiratory efforts is confirmed by the observation of Aiken and Smith (1953) on pneumothorax after block dissection of the neck. They

bronchus, leads to a leak of air into the lung parenchyma. Tracking up along the pulmonic vessels to the hilum, it enters the mediastinum and ruptures into the pleural sacs through the same area of mediastinal pleura below the position of the thymus. This has been demonstrated in the experimental work of Macklin and in numerous clinical observations, for instance those of Flett (1944) and Graebner (1939).

"The child lay with the head and neck markedly extended."

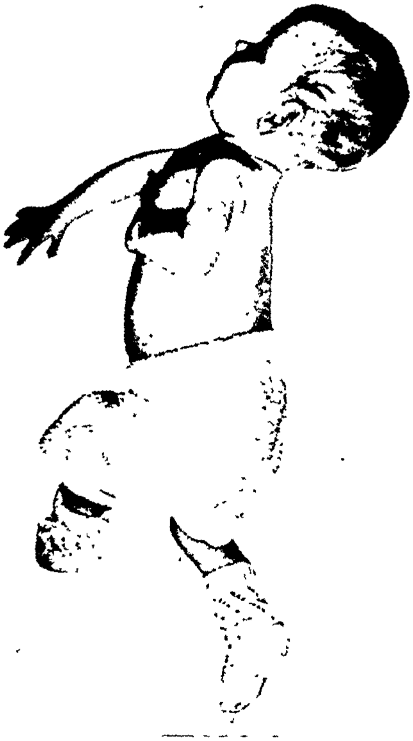

Fig. 6.

Extension of the neck produced by tracheal compression by tuberculous glands.

This attitude, so suggestive of meningitis, is also adopted by infants suffering from apical pneumonia. The photograph (Fig. 6) is of a child who, suffering from tuberculous mastoiditis, developed retraction of the neck to such a degree that tuberculous meningitis was strongly suspected. The cause, however, proved to be a mass of tuberculous glands pressing on the trachea and slightly narrowing it, as was confirmed later by bronchoscopy. The child eventually made a complete recovery.

Pædiatricians are aware that an infant, suffering from retropharyngeal abscess, tends to adopt this attitude, and over several years the author has noticed it in infants who have partial upper respiratory obstruction.

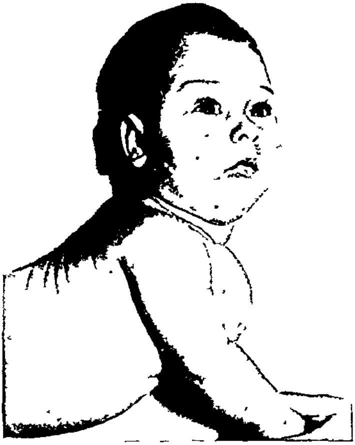

FIG 7

Extension of neck produced by vallecular fossa cyst

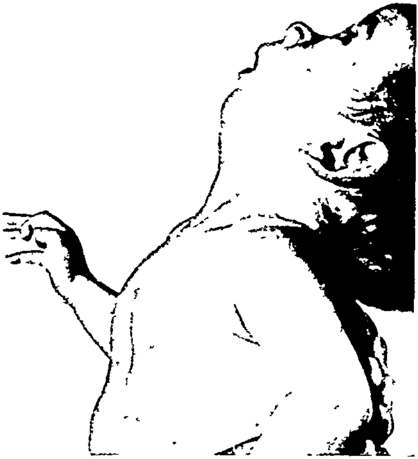

FIG 8

Extension of head and neck in Pierre-Robin syndrome

Two instances are shown. The child (Fig. 7) had a cyst of the vallecular fossa, which had thrust the epiglottis down over the glottis. Respiratory embarrassment was slight but definite. The child in Fig. 8 was afflicted with the Pierre-Robin syndrome, and there was a certain degree of interference with normal respiration from the mal-development of the lower jaw and the root of the tongue.

It appears that certain infants will adopt this position if the airway is inadequate, and it is suggested that hyper-extension of the neck after tracheostomy produces distortion of the normal position of the tube and so may lead to sub-glottic stenosis.

"The airway through the glottis proved insufficient to permit normal breathing."

The sub-glottic tissues are quick to react by swelling and subsequent scarring to injury, such as too high an opening of the trachea. The use of a stabbing incision into the trachea has also been blamed as a cause of sub-glottic stenosis: a dull blade or a resistant patch of cartilage may cause the anterior tracheal wall to bend inwards, a deformity which may be exaggerated and perpetuated by thrusting the cannula through a narrow, tight opening. The surgeon who performed the tracheostomy on the child under review was well aware of these dangers and, having incised the trachea transversely between the third and fourth rings, had, when normal respiration was established along the blades of dilator forceps, excised a small piece of the third tracheal ring, to produce a round orifice of ample size to allow the easy introduction of the cannula.

The post-mortem specimen showed that the upper two tracheal rings had in fact been buckled back to form a transverse ridge across the sub-glottic region. This was undoubtedly the cause of the failure to decannulate permanently, but why had the buckling occurred? The clue to the problem lies in the presence of the traumatic erosion in the anterior face of the trachea at a lower level. This could have been produced only by the pressure of the lower end of the cannula; indeed the attending housemen and nurses had remarked on changing the tube some weeks before the child's death, that its lower end seemed to hitch on something lower down the trachea.

For the lower end of the cannula to impinge so forcibly on the anterior tracheal wall as to cause an erosion, either the tube as a whole must have been too large or excessively curved, which it was not, or the upper end of the tube had to be tilted strongly backwards. The tube employed was a standard model, not too large for the infant's trachea, and its curve was something under a right angle. The radiograph (Fig. 9)—a portable taken of a struggling child must have some imperfections—shows that in fact the persistent extension of the child's neck had produced a backward tilt of the tube. The superior surface of the convexity of the tube had in consequence pressed against the intact first and second tracheal rings and so buckled them back into the lumen of the upper trachea. The tilt of the tube had caused the lower end to dig into the anterior surface of the trachea at a lower level, so producing the traumatic ulcer.

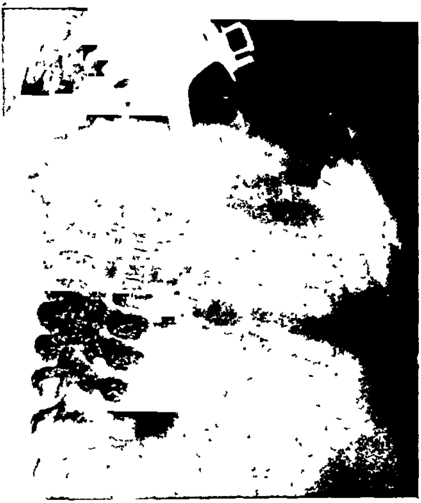

FIG 9
Neck of tracheotomized infant, showing extension

It is suggested that this peculiar mechanism of extension of the neck may be more common among infants suffering from air hunger than reference to it in the literature would seem to indicate. Its appearance during the first few days after tracheostomy in response to partial obstruction of the tube lumen may be responsible for tilting the upper end of the tube backwards and so bending the first ring in. Even if the posture is maintained for only a brief period, the cartilage may remain deformed, and so lead to permanent sub-glottic stenosis.

In conclusion the author would repeat Wilks's modest words: ". . . as we have never seen this alluded to by authors, we would speak of it with some hesitation from our small data."

Summary

1. The case history of an infant who died some time after tracheostomy is reviewed in detail.

2. Tension pneumothorax occurred and its causation is reviewed. Suggestions are made for its prevention.

3. Extension of the neck in infants, who are suffering from partial obstruction of the upper air passages, is described, with characteristic case histories.

4. The post-mortem findings in this unsuccessful case are examined and found to have been caused by persistent extension of the neck in the post-operative period.

5. A mechanism is suggested whereby sub-glottic stenosis may occur after tracheostomy.

Acknowledgments

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MUCOUS MEMBRANE PEMPHIGUS

By A G GIBB (Dundee)

PEMPHIGUS affecting the mucous membranes of the mouth and throat is a rare disease. At times it may pass undiagnosed especially if there are no associated lesions in the skin or mucous membranes elsewhere in the body.

Classification

The exact relationship of the various forms of pemphigus is still in doubt, but the following classification, based on the histological appearances of the lesion, was suggested by Lever (1953).

True Pemphigus (Intra-epidermal bulla)	Pemphigoid (Sub-epidermal bulla)
1 Pemphigus vulgaris malignus (Acute pemphigus vulgaris) Pemphigus vegetans	1 Bullous Pemphigoid (Chronic pemphigus vulgaris)
2 Pemphigus foliaceus Pemphigus erythematosus	2 [illegible]

Incidence of Mouth and Throat Lesions

The mucous membranes of the mouth and throat are commonly involved both in acute pemphigus vulgaris (nearly 100 per cent.) and in ocular pemphigus (90 per cent.) but are much less frequently affected in pemphigus foliaceus (7 per cent.) and in bullous pemphigoid (34 per cent.) (Lever, 1953).

Costello *et al.* (1957) have drawn attention to the high incidence of cases where the disease starts in the mouth (55 per cent.), in these cases the oral lesions may precede cutaneous or other mucosal lesions by weeks, months or occasionally years. They felt that dentists and laryngologists should be constantly on the look-out for primary pemphigus lesions in all cases of bucco-pharyngeal ulceration, and stressed the importance of early diagnosis and prompt treatment with corticosteroid drugs.

Primary oral lesions are, however, extremely rare in pemphigus foliaceus and bullous pemphigoid and these conditions will not be considered further.

Histology

Acute pemphigus vulgaris and ocular pemphigus can be separated histologically by examination of intact bullae; early diagnosis is essential as both groups differ greatly in clinical features, prognosis and complications. The histological differentiation is the result of the work of Civatte (1943). He discovered that the lesions of pemphigus vulgaris show degenerative changes in the epidermis leading to a disappearance of the intercellular partitions; to this he gave the name acantholysis. The cellular break-down leads to the formation of intra-epidermal clefts which enlarge to form bullae. These normally form immediately superficial to the basal layer of the epidermis (Fig. 1). Detached epidermal cells showing degenerative changes lie free in the cavity of the bullae.

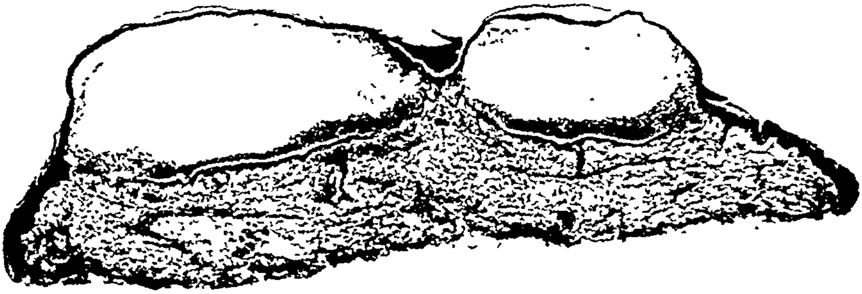

FIG. 1.
Pemphigus vulgaris: intra-epidermal bullæ with splitting of the layers of the epidermis. (Case I)

In ocular pemphigus, all changes are sub-epidermal. The bullae result in detachment of the entire epidermis from the underlying corium; the epidermis shows no degenerative changes and acantholysis is absent (Fig. 2). Some observers have applied the name pemphigoid to this condition as they do not regard these changes as characteristic of true pemphigus.

Clinical Features

Pemphigus vulgaris

Pemphigus vulgaris commonly affects those in middle age.

Skin Lesions

The skin lesions are fairly typical, the epidermis tending to slide off in large sheets (with or without the formation of bullae), leaving painful denuded areas which tend to enlarge rather than heal. Lesions are frequently present around the lips and nose.

Mucosal Lesions

Although mouth ulcers are at least as common as conjunctival lesions, the disease is seldom diagnosed until it attacks the conjunctiva —hence the name ocular pemphigus. Bullae lead to sub-epidermal scarring which is a marked feature of the disease. The lesions are, as a rule, painless and give little trouble until the effects of scarring are evident. The *eye lesions* start with a simple catarrhal conjunctivitis associated with mucoid discharge and sometimes a burning or itching sensation. Bullae are seldom seen but sub-conjunctival scarring takes place, leading to shrinkage of the conjunctiva. Adhesions form between the palpebral and bulbar conjunctiva eventually obliterating the fornices and fixing the lids to the globe. The upper and lower lids may become adherent with narrowing of the palpebral fissure. Entropion and xerosis follow and lead to corneal ulceration and blindness. The *mouth lesions* tend to be painless. Sometimes bullae are seen but more frequently indolent denuded areas which heal slowly eventually giving rise to white scars. The scars may result in adhesions; sometimes these form between the buccal mucosa and alveolus obliterating the buccal sulcus; sometimes they involve the soft palate, pillars of fauces and uvula. Hoarseness may result from bands of adhesions in the *larynx*. Fibrous stricture of the *œsophagus* may occur as a rare complication; nine such cases were traced by Lever (1953). If the *nose* is involved, chronic inflammation and erosions may eventually result in fibrous adhesions and in atrophic rhinitis. The *genital* region is frequently involved; bullae and erosions occur on the glans penis or vulva and vagina; these are followed by scarring, and vaginal atresia has been known to occur. The *anal* mucosa is less commonly affected.

Skin Lesions

The skin is involved in approximately 50 per cent. of cases. Usually the regions of the eyelids, face, head and neck are affected. Skin lesions may precede mucosal lesions by several years.

The disease is practically never fatal unless severe œsophageal stenosis is present.

Treatment is often ineffectual as the diagnosis is seldom made until scarring has taken place. Church and Sneddon (1953) found all forms of treatment of little value. Corticosteroid therapy does, however, have a good effect on active erosions, but has no effect on adhesions and strictures in long-standing cases.

Case Records

The following three cases illustrate the main features of both types of pemphigus.

Case I Mrs S , aet 52 (a case of acute pemphigus vulgaris), had eighteen dental extractions under local anæsthesia for painful and bleeding gums with halitosis The gums healed but 3–4 weeks after the extractions the mouth and throat became increasingly painful and there was marked pain on swallowing Gross ulceration was present on the lower lip, floor of mouth, both cheeks and the pillars of fauces The ulcers showed a tendency to bleed Two months later red papules appeared on the abdomen and back These gradually enlarged and became vesicular The vesicles ruptured leaving areas of weeping denuded epithelium The process spread and large areas appeared on the trunk and limbs Bullae were also seen in the mouth but they ruptured very rapidly giving rise to superficial erosions Epistaxis and hoarseness indicated involvement of the nose and larynx She was later referred to the Ear, Nose and Throat Department On examination, I noted areas of denuded epithelium on the left side of nose, buccal mucosa hard and soft palates, epiglottis and left vocal cord Microscopic examination of a bulla showed changes typical of pemphigus vulgaris, with splitting of the deepest layers of the epidermis and acantholysis The skin lesions cleared up after treatment with ACTH, but, as frequently happens, the mucosal lesions persisted These lesions are now no longer painful

Case II Miss L C , aet 65, (a case of ocular pemphigus) reported because of itchiness of the right eye She was kept under observation and the lower fornix gradually became obliterated The diagnosis of ocular pemphigus was made and other mucous membranes were examined in detail She was found to have painless erosions on the left side of nasal septum, buccal epithelium, and palate She also had an adhesion across the right vallecula and a healed ulcer on the aryepiglottic fold She gave a history of genital erosions for 30 years and examination of the genital area showed superficial ulcers and bullae on the vulva Repeated examinations of the mouth and throat revealed bullae on the buccal epithelium and palate on several occasions

Treatment by cortisone has helped to keep the eye condition under control

Case III A McK Male, aet 69 (a case of ocular pemphigus), first reported to the Eye Department and was treated for "chronic conjunctivitis' When, five years later, he developed obliteration of the fornices in the left eye, the diagnosis was altered to "essential shrinkage of conjunctiva caused by ocular pemphigus" Progressive contracture caused entropion which led to damage to the cornea from the eyelashes Three years later the lids became completely adherent to the eyeball and severe chronic ulceration with pain and blindness resulted, evisceration was carried out In the meantime, a similar condition was developing in the right eye and progressive blindness followed

About this time he was referred to the Ear, Nose and Throat Department and was found to have a left-sided atrophic rhinitis and an obscure chronic pharyngeal condition, with the appearance of a membrane on the posterior pharyngeal wall, epiglottis, valleculae, and false cords. The condition was diagnosed (after repeated examinations and biopsies) as a chronic non-specific pharyngitis, together with an atrophic rhinitis. At a later date, contractures of the aryepiglottic folds pulled the epiglottis backwards and downwards giving rise to stridor. Seven years later the man was sent back to the department because of severe dysphagia. He still had active superficial ulceration of the buccal and pharyngeal mucosa. The pharyngeal condition had, however, advanced greatly for the palate was now adherent to the posterior pharyngeal wall and the nasopharynx seemed entirely closed off. Radiological examination, however, using lipoidol, showed that a small communication did in fact exist (Fig. 3). A barium swallow showed that the dysphagia was due to a

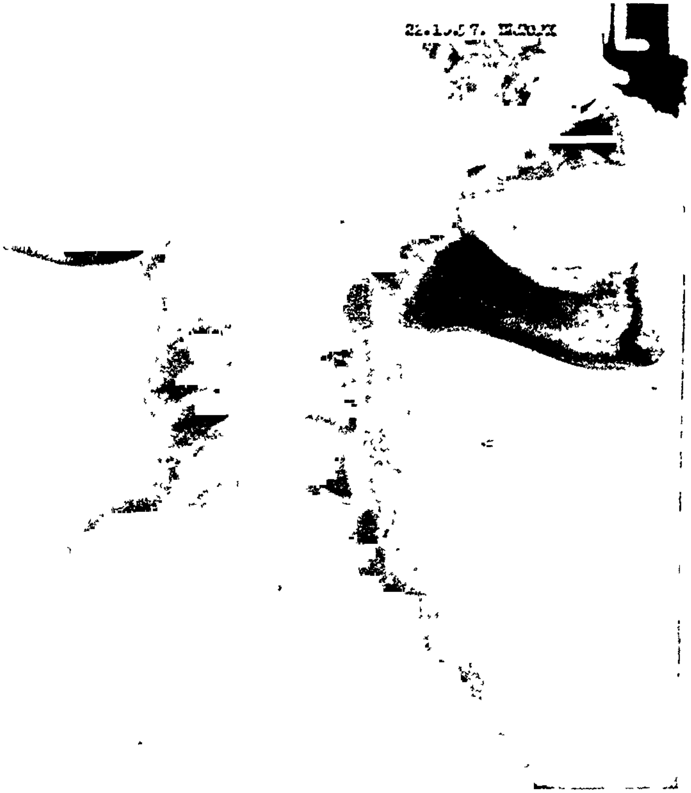

Fig. 3

Radiograph with lipiodol showing gross narrowing of nasopharynx due to ocular pemphigus. (Case III.)

œsophageal stricture had been successfully dilated by the passage of bougies. Two large peptic ulcers were present in the stomach.

Conclusion

Two varieties of pemphigus are likely to affect the mucous membranes of the mouth and throat. These are:

1. Acute pemphigus vulgaris—a serious disease which carried a mortality of 90 per cent. before the introduction of corticosteroid therapy. This disease affects the middle-aged and is associated with painful mouth erosions which show little tendency to healing or fibrosis; bullae are seldom seen.

2. Ocular pemphigus—a chronic disease generally found in older people, seldom if ever fatal, but causing progressive blindness from shrinkage of conjunctiva. Other mucous membranes are invariably affected and bullae may be seen in the mouth and throat. These rupture and give rise to painless erosions which heal slowly with the formation of dense scar tissue leading to fibrous contractures many years afterwards.

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tight upper œsophageal stricture (Fig. 4); œsophagoscopy revealed active erosion of mucosa immediately above the stricture. It now became apparent that the nose and throat lesions were local manifestations of ocular pemphigus. Further investigations revealed Paget's disease affecting skull and long bones.

FIG 4

Barium swallow outlining severe upper œsophageal stricture caused by ocular pemphigus (Case III)

Treatment by cortisone and ACTH brought improvement in the throat lesions, but, at the end of the treatment, the patient developed broncho-pneumonia and died.

Post-mortem examination revealed, in addition to the pharyngeal erosions, an almost complete obliteration of the nasopharynx. The œsophageal stricture had been successfully dilated by the passage of bougies. Two large peptic ulcers were present in the stomach.

Conclusion

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1. Acute pemphigus vulgaris—a serious disease which carried a mortality of 90 per cent. before the introduction of corticosteroid therapy. This disease affects the middle-aged and is associated with painful mouth erosions which show little tendency to healing or fibrosis; bullae are seldom seen.

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until the incision is healed, when it is done fortnightly, then monthly, then three-monthly and lastly at six-monthly intervals All the results shown here have been worked out from the latest audiogram

In all, 146 mobilization operations were performed on them in this series One hundred and twenty-two individuals were operated on of which 14 had both ears done Ten of the cases which failed were re-mobilized, and over and above this, 2 of the failures were fenestrated

Broadly speaking, when I started this series of cases there were very few guiding factors as to what type of case would be successful, and in view of this, all cases of otosclerosis, however advanced, who expressed a desire for operation were accepted Several of these cases I would never accept for fenestration and although some of these have been failures and I would not dream of attempting them again, others, surprisingly enough have had very good results and are pleased with them even though their hearing has not come up to what is usually regarded as a socially adequate level

Table I shows the breakdown of these results on all cases in decibel increases The average decibel increase in all cases has been obtained by adding the gains or losses over the frequencies of 250, 500, 1 000, 2,000 and 4,000 c p s and taking the average

TABLE I

No of patients	122
No of cases with both ears operated on	14
No of failures re mobilized	10
Total	146
No of failures fenestrated	2

Average Increase over 250–4 000 c p s	No of Cases	%
40 db and over	9	24
30 – 39 db	12	
20 – 29 db	14	
15 – 19 db	14	9 6
10 – 14 db	18	12 3
5 – 9 db	26	17 8
0 4 db	24	16 4
–1 – –9 db	14	11 6
–10 or over	3	
	134	
No of failures re mobilized or fenestrated	12	
	146	

From this it can be seen that the total average increase over all cases was 9 7 db and that

24 per cent. of all cases showed an average increase of over 20 db.
33·6 per cent. of all cases showed an average increase of over 15 db.
45·9 per cent. of all cases showed an average increase of over 10 db.
63·7 per cent. of all cases showed an average increase of over 5 db.
28 per cent. of all cases showed no change or a loss.

Further breakdown of the figures shows that 55 of the ears operated on were male and 91 were female.

Table II shows that sex had no appreciable affect on the success or failure of the operation.

TABLE II

Average Increase over 250–4,000 c.p.s.	Males	Females
40 db and over	3	5
30–39 db	5	8
20–29 db	6	8
10–19 db	12	20
0– 9 db	17	33
Loss	12	17
	55	91

As has been stated, no attempt was made initially to classify these cases into Shambaugh's classification. However, since then they have been allotted into their appropriate groups and Table III shows the results. I would like to stress the fact that under the heading Class C are several cases who are really not C cases at all but are cases I would not consider suitable for fenestration at all.

TABLE III

Average Increase over 250–4,000 c.p.s.	Shambaugh's Class A		Shambaugh's Class B		Shambaugh's Class C	
	No.	%	No.	%	No.	%
40 db and over	0	23·5	3	21·6	5	26·3
30–39 db	2		4		7	
20–29 db	2		4		8	
10–19 db	2	11·8	13	25·5	17	22·4
0– 9 db	9	65	14	53	25	51·3
Loss	2		13		14	
+2 failures fenestrated	2					
	19		51		76	

From this it appears that Class A has the greatest per cent. of failures but that those cases with an average improvement of 20 db. or more does not vary significantly in any of the groups.

Lastly the cases have been classified according to age.

TABLE IV

Average Decibel Increase over 250–4 000 c p s	Ages				
	10–19	20–29	30 39	40–49	50–59
40 db and over	0	2	3	2	1
30–39 db	2	2	5	4	0
20–29 db	2	5	5	2	0
10–19 db	5	4	12	5	2
0– 9 db	3	11	16	16	8
Loss	3	8	11	7	0

To me the interest in this table is the comparative success of the under 20's (in these cases the initial loss and therefore subsequent gain is minimal) and the comparative failures of the over 50's

Complications

The complications of the operation are singularly few

They may be divided into
(*a*) Immediate
(*b*) Remote

(a) *Immediate*

(1) *Facial palsy* Two cases have been encountered, presumably due to the spread of the xylocaine used as a local anæsthetic as they have only lasted for about an hour and then disappeared

(2) *Perforation of the drum* This has occurred on several occasions during the incision and separation of the flap At the end of the procedure and after replacing the flap the clot which invariably lodges in the antero-inferior part of the external auditory meatus during the operation is pulled out and placed over the perforation which it invariably seals No permanent perforation has been observed as a result of this injury, but it is one to be avoided as I feel strongly that the final gain is never as high as the initial gain would lead one to hope

(3) *Section of the chorda tympani nerve* This has occurred either inadvertently when retracting the annulus fibrosus from its sulcus or deliberately to obtain a better view of the footplate Both these circumstances occurred in the earlier cases and resulted in aberrances in the sense of taste which were complained of to a greater or lesser degree but by none for long

(4) *Vertigo* This is complained of at the moment of mobilization in the vast majority of cases but is very seldom more than momentary A few cases have complained of it for a week or so but it has rapidly settled down under Avomine 15 mg t i d

(b) *Remote Complications*

(1) *Infection.* Altogether eleven cases have developed infection of the middle ear. Six of these developed it soon after the operation while still in hospital. The other five developed it after discharge following a cold during the period of healing. Four out of the eleven cases were left with a perforation, but all ears are dry. Not in all of these cases was the hearing gain destroyed as can be seen from Audiograph No. 1. The subsequent history of two of these eleven cases is dealt with later on under the heading of re-mobilization.

(2) *Trauma.* One case, in spite of warning, blew his nose vigorously in the post-operative period and detached the flap. He now has a perforation and has been a failure.

(3) *Asian 'flu and coryza.* One case lost her initial gain after an attack of Asian 'flu and was re-mobilized successfully. See Audiographs 2 and 3.

Another case lost hers suddenly after a cold and is awaiting re-mobilization.

Causes of Failure

Besides those of the complications listed above which may cause failure, the main causes are I feel:

(*a*) Eustachian Blockage.
(*b*) Failure to mobilize the stapes.

(a) *Eustachian Blockage*

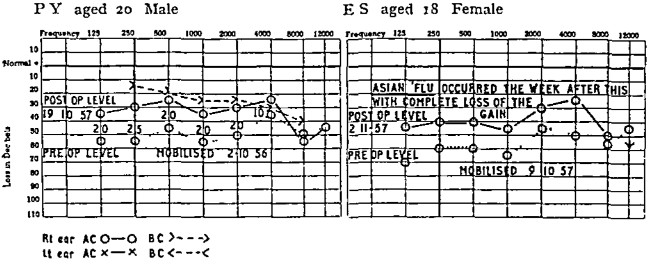
Four cases who had an immediate gain on the operating table, lost it straight away on replacement of the flap. Eustachian catheterization after healing showed the Eustachian tube to be blocked.

(b) *Failure to mobilize the stapes*

I feel that failure to mobilize the stapes adequately or at all is the greatest single cause of failure of the operation. The best cases are those in which the stapes mobilizes easily and after this, on testing, is seen to be freely mobile. The typical free bobbing motion of the mobile stapes dipping like a fisherman's float on the water is quite characteristic and pathognomonic of a good result. Equally characteristic in my series is the appearance of the tendon of the stapedius muscle which in the successful case can be seen to move with the stapes but retains fully its tone. If seen to be twisted it is highly suggestive of fracture of the crura.

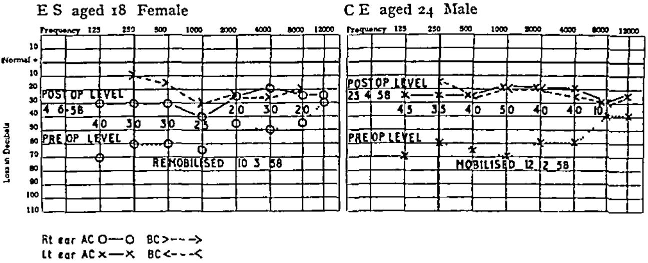
In other cases the stapes can be seen to be mobile but not freely mobile. On pressure it moves sluggishly like a door with rusty hinges. In these cases, although improvement is the general rule it is by no means good.

Lastly, there are the cases in which no amount of manœuvring will move the footplate. In the earlier cases of the series I was often bemused by the apparent mobility of some of these stapes without improvement in hearing. I realize now that I was dealing with an immobile footplate and fractured crura. As time has gone on and I have got bolder, I have mobilized more of these cases but the hard core still remains. In two of these cases I have fenestrated the footplate with immediate gain in the hearing but no lasting success. In this, as in all things, results improve with practice as can be seen from Table V below. Unfortunately, practice in this particular stage can only be obtained on the patient. Cadaver surgery gives extremely valuable experience in the surgery of the access to the stapes but none whatsoever of any value in actual mobilization. It is impossible from the mobile stapes of the cadaver to get any idea of how to mobilize the fixed stapes of the otosclerotic, especially as the mode of *fixation is so infinitely variable.*


P Y aged 20 Male

E S aged 18 Female

Figs 1 and 2.


E S aged 18 Female

C E aged 24 Male

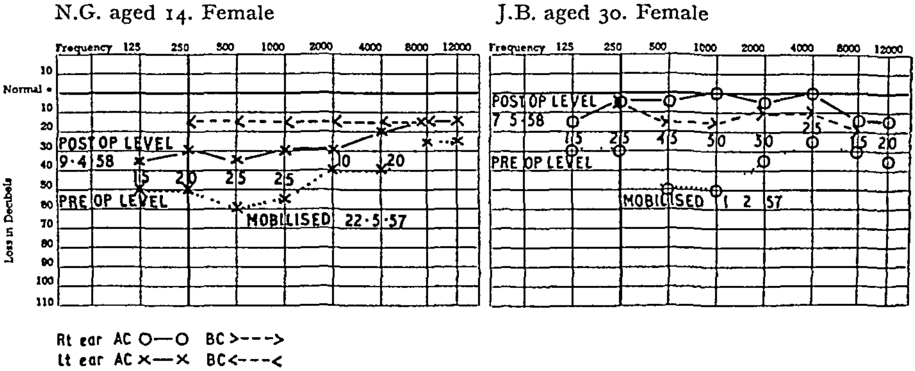
Figs 3 and 4

TABLE V

Average Increase over 250–4,000 c p s	1st 40 Cases		2nd 40 Cases		3rd 40 Cases	
	No	%	No	%	No.	%
40 db and over	1	12·3	3	23·4	5	37·5
30–39 db	2		2		8	
20–29 db	3		6		5	
10–19 db	10	20·4	12	20·5	10	20·8
0– 9 db	19	38·8	15	31·9	16	33·3
Loss	10	20·4	6	10·25	2	4·2
Failures re-mobilized	4		3		2	
	49		47		48	

As can be seen from the table the percentage of results with an average gain of over 20 db. rises from 12·3 per cent. in the first 40 cases to 37·5 per cent. in the last 40 and over 10 db. gain from 32·7 per cent. to 58·3 per cent.

Disruption of the ossicular chain. This occurs on occasions, chiefly at the incudo-stapedial joint. If the incus and stapes are put into contact in their usual anatomical positions, this does not give rise to failure of the operation. Occasionally even complete disruption may not give rise to a failure. For instance, in one case great resistance was felt at the footplate on attempted mobilization. Pressure was gradually increased until eventually it suddenly mobilized with a noise that can only be described as a "Clang". The crura were seen to fly across the operative field and were ultimately recovered from the hypotympanum, with a torn piece of stapedius tendon attached to them. The patient, a phlegmatic young man, was heard to remark that that was one "——" of a noise but that he could now hear. His ultimate average increase in


N.G. aged 14. Female

J.B. aged 30. Female

FIGS. 5 and 6.

hearing was 41 db (Audiograph No 4) Although the result was gratifying I hope personally never to see another case like it

Re-mobilization of the Stapes

Ten of these cases were re-mobilized Their initial distribution was as follows

TABLE VI

Causes of Failure	1st 40 Cases	2nd 40 Cases	3rd 40 Cases
Failure to mobilize	4	2	
Infection		1	1
Asian flu			1
Neglect on part of patient during healing period			1
Total	4	3	3

Re-mobilization, in all these cases except two, presented no difficulties other than the presence of a few adhesions The results of the eight cases which presented no difficulties are shown in Table VII below and the other two are considered separately

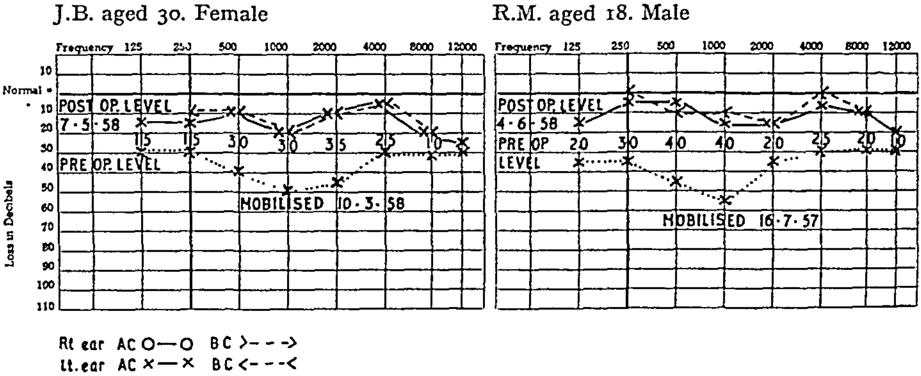
TABLE VII

Average Increase over 250–4 000 c p s	Re mobilized in 1st 40 Cases	Re mobilized in 2nd 40 Cases	Re mobilized in 3rd 40 Cases
40 db and over	0	0	0
30–39 db	1	0	1
20–29 db	1	1	0
10–19 db	1	1	1
0– 9 db	1	0	0
Loss	0	0	0
	4	2	2

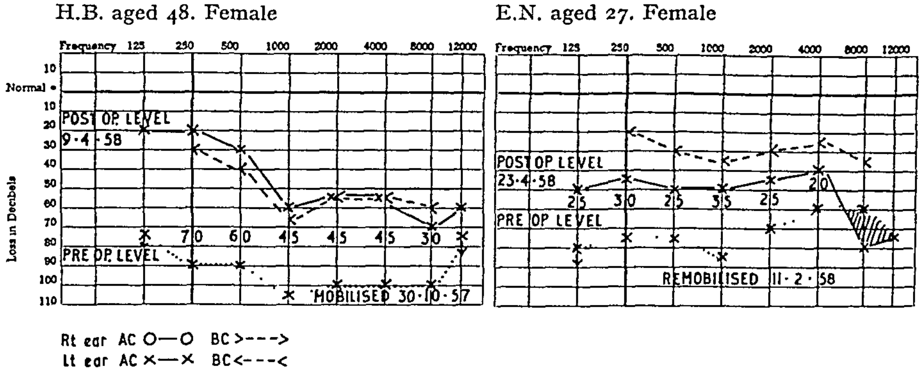
The other two cases were situated in the 2nd and 3rd 40 cases respectively They developed an acute suppurative otitis media in the operated ear after discharge from hospital Their ears dried up but they were both left with posterior marginal perforations

The one in the 2nd 40 was re-admitted, re-mobilized and a myringoplasty performed Her hearing came up nicely to the clinical tests and she was discharged On the night of her discharge she went to a dance, caught a cold, developed another infection with otorrhœa and reverted to her pre-operative level

The second case (in the 3rd 40) had a fair initial gain but caught a cold three weeks after discharge, followed by acute suppurative otitis media. When this had healed she was re-admitted and re-mobilized and a myringoplasty performed. At operation it was seen that the long process of incus had been destroyed by the infection. Her ultimate average gain was 10 db. and is still rising.



FIGS. 7 and 8.



FIGS. 9 and 10.

Discussion

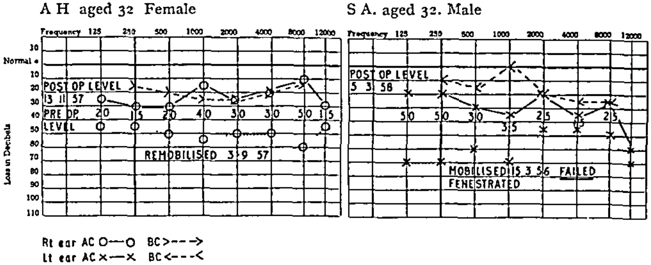
I think from these figures that patients can be fairly offered a 50 per cent. chance of a significant improvement in their hearing and a 30 per cent. chance of a substantial improvement, and that this can be offered to all patients suitable for fenestration and to a large number who are unsuitable for it.

There remains, however, the very large block of cases (34·2 per cent.) who have an improvement of only 0–9 db. In Table I I have broken these into two categories.

(*a*) 0–4 db. (16·4 per cent.)
(*b*) 5–9 db. (17·8 per cent.)

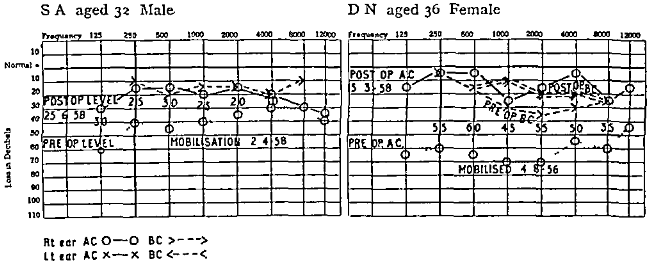
Those in group (*a*) must be written off as failures although they are no worse.

Those in group (*b*) are not entirely so. A lot are very satisfied with their results and will not consider an aid. People who work in our noisy Northern industry, particularly the women are often proficient lip readers, even when not deaf. Otosclerotics as a rule are even more proficient. An average increase of 5–9 db. will often, in these cases make the difference between social adequacy and loneliness.


A H aged 32 Female

S A. aged 32. Male

FIGS 11 and 12


S A aged 32 Male

D N aged 36 Female

FIGS 13 and 14

In spite of this a constant group of indifferent results can only be a source of worry to any surgeon, especially as, in this series, it is the only constant figure in the three sets of 40 cases. I can only hope that with time, practice and knowledge this figure will diminish.

Lastly, a few selected audiographs of the cases:

(1) The youngest case of the series.
N.G. Age 14 years. Female. Mobilized 22.5.57. Average gain 20 db.
Audiogram No. 5.

(2) Two cases where the hearing has returned to normal.
J.B. Age 30 years. Female: Right ear mobilized 1.2.57. Average gain 35 db.; Left ear mobilized 10.3.58. Average gain 27 db.
Audiogram Nos. 6 and 7.

(3) R.M. Age 18 years. Male. Mobilized 16.7.57. Average gain 31 db.
Audiogram No. 8.

(4) Greatest average gain.
H.B. Age 48 years. Female. Mobilized 30.10.57. Average gain 53 db.
Audiogram No. 9.

(5) Two re-mobilizations.
E.N. Age 27 years. Female. Re-mobilized 11.2.58. Average gain 27 db.
Audiogram No. 10.
A.H. Age 32 years. Female. Re-mobilized 3.9.57. Average gain 27 db.
Audiogram No. 11.

(6) An initial failure in one ear followed by fenestration and later by mobilization of the other ear.
S.A. Age 32 years. Male. Left ear mobilized 15.3.56. Failed. Fenestrated. Average gain 31 db.; Right ear mobilized 2.4.58. Average gain 22 db.
Audiogram No. 12.

(7) The only case in the series where in my opinion there has been a significant gain in the bone conduction.
D.N. Age 36 years. Female. Mobilized 7.8.56. Average gain 53 db.
In this case the bone conduction gained 20 db. at 1,000 c.p.s.
15 db. at 2,000 c.p.s.
10 db. at 4,000 c.p.s.
Audiogram No. 14.

Acknowledgments

In conclusion, I would like to thank those of my colleagues at the Manchester Ear Hospital who have referred some of these cases to me, and to Mr. Denzil Brookes, B.Sc., Physicist-in-Charge, The Hardman Street Hearing Aid Clinic who has been responsible for all the audiometric surveys of the series.

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ORAL PREDNISONE IN NASAL ALLERGY

By H J SHAW and B P SCRIVENER (London)

In this study 28 patients with allergic disease of the nasal mucous membrane have been treated with oral prednisone The primary condition in most cases was nasal allergy, but in some a mixed infective element predominated

Before considering the results obtained, it is necessary to review some fundamentals of immunology and physiology upon which this type of treatment is based

Following the original work in 1905 by Von Pirquet and Schick on serum sickness, Von Pirquet (1906) defined allergy as "an acquired specific capacity to react" Upon this concept is based the present day cellular theory of allergy, which postulates that the allergic phenomena observed clinically are due to the release of histamine and other substances from sensitized cells These cells have become sensitized by the attachment to them of antibodies produced by the host from previous exposure to the antigen The antigen-antibody reaction occurs in these cells and disrupts them, the histamine and other substances then liberated cause an inflammatory reaction in the affected tissues, which is histologically very similar to that resulting from damage to the tissues caused by a "primary irritant" such as bacterial toxin, and by many chemicals

This can be said to be the typical allergic response in nasal mucous membrane, but in some organs the antigen antibody reaction may cause a contraction of smooth muscle rather than an inflammatory reaction, in the lungs bronchospasm may result or in the gut increased peristalsis

Methods of Control

For many years now, attempts at curative treatment for allergic rhinitis have been based on the proved immunological concept of specific desensitization Gradually increasing doses of the responsible antigen are injected in an attempt to saturate all the available antibodies, and if this is successful, no reaction occurs when further antigen is presented

On the other hand the antigen-antibody reaction has been attacked with non-specific measures, whose results, though by definition palliative, are much more rapidly apparent The first of these were the antihistamines, which McGovern (1956) has shown to increase the density and toughness of the hyaline layer covering the vascular endothelium, and of the intercellular cement substance. In this way, vascular permeability is decreased, and the changes seen in an inflammatory reaction are inhibited.

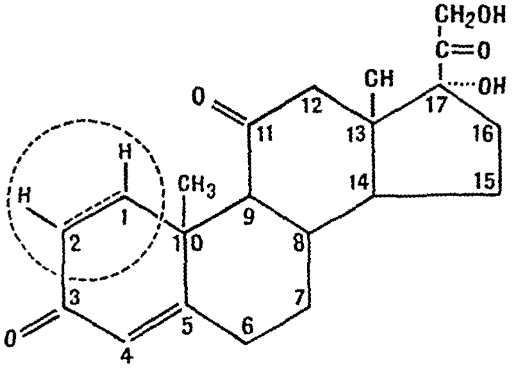
More recently, adrenal steroids and ACTH have been used because of their powerful anti-inflammatory action. The precise mechanism is unknown, but it is likely that the antigen-antibody reaction is inhibited to some extent, that vascular permeability is decreased, and that the action of the spreading factor hyaluronidase is opposed. The release of histamine and associated substances does not in fact appear to be affected.

The end result is that adrenal steroids suppress an inflammatory reaction whatever its cause, and this fact must be borne in mind when these substances are used therapeutically. The humoral and cellular responses occurring in an inflammatory process due to bacterial invasion are the chief mechanisms for keeping the invaders localized, and rapid dissemination throughout the body may occur if these responses are suppressed.

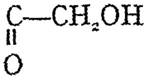
The fact that some favourable therapeutic response has been obtained in three primarily infective cases and in one allergic case with marked super-added infection in the present series may also be accounted for by the non-specific anti-inflammatory action of adrenal steroids.

Adrenal Steroid Physiology

All the cortical hormones, of which at least 28 have now been isolated, consist of 4 benzene rings arranged as shown below in the structural formula of prednisone.



Prednisone

The steroids having physiological activity possess a double bond between C4 and C5, a lactonic oxygen at C3, and a  $\underset{\underset{\displaystyle O}{\|}}{C}\text{—}CH_2OH$ side chain at C17.

The chief function of these hormones is the control of salt and sugar metabolism in the body, although they also exert an important influence on sexual function Those responsible for the first two functions represent the mineralocorticoids and glucocorticoids of Selye (1949)

The mineralocorticoids are probably formed in the zona glomerulosa of the adrenal cortex Their principal representative is aldosterone, which promotes the excretion of potassium and the retention of sodium and chloride, and therefore of water, by the kidney Hypertension may result from its overaction

The sex hormones, thought to be formed in the zona reticularis, are chiefly androgens, but oestrogens and progesterone are also produced

The sugar hormones, formed in the zona fasciculata, include cortisone and hydrocortisone, having at the C11 position a ketonic oxygen and hydroxyl group, respectively They oppose the action of insulin, promote protein metabolism and the breakdown of liver glycogen to glucose, and so tend to raise the blood sugar They also possess the anti-inflammatory properties which are the chief interest of this study

The production of aldosterone is largely autonomous, whereas the production of the sex and sugar hormones is controlled by ACTH produced in the adenohypophysis The secretion of ACTH is controlled in turn, by the blood level of the sex and sugar hormones Sayers (1955) believes that this is done by a direct mechanism, determined by the hormone levels in the blood reaching the adenohypophysis However, Harris (1944, 1948) has advanced convincing experimental evidence of a *portal system of vessels connecting the hypothalamus and the adenohypophysis* He believes that falls in the steroid level of the blood reaching the hypothalamus cause chemical mediators to be elaborated there, and that these are carried to the adenohypophysis in the vessels of the portal system, so causing the liberation of ACTH

Thus, one of the effects of the therapeutic use of adrenal steroids is the depression of ACTH production by the adenohypophysis, with secondary atrophy of the adrenal cortex This serious endocrine disturbance must always be borne in mind whenever a long course of treatment is planned

Clinical Action and Application of Adrenal Steroids

Prednisone was introduced into clinical use in 1954 Its formula is identical with that of cortisone, except that it has an additional double bond in the first ring between C1 and C2 This is also the difference between prednisone and hydrocortisone Both synthetic analogues have properties similar to those of the naturally occurring substances, except that they have slightly less mineralocorticoid action than the latter The synthetic substances are 4 to 5 times more active, weight for weight, and some clinical conditions have responded to these newer substances, when cortisone and hydrocortisone have been ineffective.

It is interesting to note the profound effect on potency exerted by these minor changes in the structural formula. For example, by the addition of a methyl group at C2 a synthetic steroid has been produced which is a much more powerful mineralocorticoid than aldosterone, previously the most powerful one known (Byrnes *et al.*, 1956). This new substance has now been shown to cause nephrosclerosis and periarteritis nodosa when injected into monkeys (Selye and Bois, 1957).

Known Side Effects

The retention of sodium, chloride and water can cause œdema, including moon face, and hypertension. Virtually symptomless peptic ulceration may occur and progress insidiously to perforation, the clinical picture being masked by the anti-inflammatory effect of the steroid.

Re-activation of tuberculous foci, mental changes and glycosuria are well recognized complications. It has also been suggested that susceptibility to sinus infection is increased (Samter, 1957). Recently, histological evidence of acute pancreatitis has been obtained at autopsies on patients who were being treated with adrenal steroids at the time of their death.

In the present series, none of the more serious side effects has occurred; however, in most cases the dosage has been small and the duration of treatment short.

Contra-indications

Unless their use is considered to be life-saving, adrenal steroids are contra-indicated where there is evidence of past or present peptic ulceration, tuberculosis or mental instability. Also, hypertension, congestive cardiac failure, chronic renal disease or any condition in which salt and water retention are undesirable usually preclude this type of treatment. Specific inquiry about the above conditions must always be made before the start of treatment.

Use of Adrenal Steroids in Diseases of the Air and Upper Food Passages

There are records of the use of these substances in the following conditions: allergic and infective disease of the nasal cavity and sinuses, C.S.O.M., otitis externa, otosclerosis, Ménière's disease, apthous ulceration of the mouth and pharynx, malignant granuloma, glottic œdema, corrosive injuries of the œsophagus, Bell's palsy, Costen's syndrome, bronchial asthma and now tuberculosis (*British Medical Journal*, 1957). They may also be used as a prophylactic measure against iodism in bronchography.

A voluminous literature on the use of steroids for such conditions has sprung up since 1950, when these substances first became available on a commercial scale In nasal allergy and infection, most reports concern their topical use, often administered with naso constrictors and antibiotics In those reported series where steroids have been used systemically, prednisone has not been commonly employed, and where prednisone has been the drug used, no clear evidence of follow-up after cessation of treatment has been found in a fairly comprehensive search of the literature (Demartini *et al*, 1955, Anderson and Ogden, 1956, Brown and Seideman, 1956, Hotchkiss, 1956, Luippold, 1956, American Academy of Allergy, 1956, Rosa *et al*, 1957)

Present Series

This series is selective only in relation to the severity of symptoms, and consists of 28 adults (Table I) All patients for whom an adequate record is available and who have been treated with oral prednisone for a nasal condition have been included There were 21 females and 7 males having a mean age of 42 years, ranging from 21 to 64 years

TABLE I

Summary of Patients Treated

Primary Nasal Condition		
Allergic rhinitis (perennial) mixed chronic infection in 12		23
Chronic suppurative sinusitis and rhinitis mixed nasal allergy in all		5
Associated Factors		
Nasal polypi	17	
Bronchial asthma	7	
C S O M	3	
Otitis externa	2	
Rhinitis medicamentosa	1	
Total patients		28

Before commencing this study in 1956, it was decided to limit acceptance of patients to those in whom all other methods of treatment had failed, and who presented the most severe symptoms and signs Most of the patients could truthfully be called veterans, having had recurrent symptoms for many years or since childhood Each had had three or more of the following forms of treatment environmental control, skin testing and specific desensitization, zinc ionization, various forms of intranasal cautery or diathermy, removal of polypi (often on numerous occasions), antihistamines cortisone snuff, hydrocortisone injections into polypi, S M R, intranasal or transantral ethmoidectomy, Caldwell-Luc antrostomy, antral lavages and hydrocortisone nasal spray Four patients had previously been given cortisone or hydrocortisone systemically from severe nasal allergic disease with oral prednisone are presented and discussed. It is concluded that the use of oral prednisone in the severe forms of nasal allergy is useful and justified.

Acknowledgments

Sincere thanks are due to Professor F. C. Ormerod and Mr. J. C. Hogg for much encouragement and permission to treat their patients. Also to Mrs. T. M. Woof for assistance with the bibliography.

Messrs. Roussel have kindly supplied all the prednisone used in this trial.

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CLINICAL RECORDS

CARCINOMA OF THE CERVICAL ŒSOPHAGUS TREATED BY END-TO-END ANASTOMOSIS

By LENNART GISSELSSON (Sweden)

THE results of treatment of cancer of the hypopharynx are not very encouraging Radiation therapy has a five-year cure rate of 10–15 per cent Owing to the radiosensitivity of the laryngeal cartilages, complications such as chondritis, are common This complication is painful and often leads to pronounced stenosis of the larynx requiring temporary or permanent tracheostomy

In the surgical treatment of cancer of the hypopharynx and the cervical œsophagus most authors claim that laryngectomy is always necessary This implies a severe trauma Shaw and Ormerod (1957), for example, write, "We would agree with Conley who states that total pharyngo-laryngectomy with primary closure by skin tube must be used when resection of the primary tumour in the hypopharynx or cervical œsophagus necessitates excision of more than 70 per cent of the mucosal circumference We also agree with Negus who states that the larynx must be removed in the surgical treatment of malignant tumours at or near the mouth of the œsophagus "

However, other authors such as Sisson (1956) stress the possibility of an end-to-end anastomosis conserving the larynx after excision of the cancer He has operated on one patient in this way but he says nothing about the result This surgical possibility appears to have received little attention, probably because the tumours are usually not detected until they have become too extensive to be removed by this method That this procedure may be useful in selected cases is, however, apparent from the following case extract of a patient who was operated upon more than 3 years ago and who still feels well

In 1954, a woman, aged 70, reported that she had for the last 6 months had swallowing difficulties Roentgen examination showed no disorder of the act of deglutition, but at the level of the seventh cervical vertebra the œsophagus was stenosed and the mucosal outline appeared stiff along a segment about 1 cm long (Fig 1) Above the stenosis the œsophagus was slightly dilated, below, it was of normal appearance Whether the narrowing was a manifestation of Paterson's syndrome or whether it was due to canceromatous infiltration was not known Œsophagoscopy was therefore performed A tumour was seen in the posterior œsophageal wall 3 cm below the mouth of the œsophagus The tumour appeared to be annular Microscopic examination of a biopsy specimen showed the characteristics of highly differentiated squamous celled carcinoma

It was decided to operate and, if necessary, to perform laryngectomy The œsophagus was approached *viâ* an incision in the right side of the neck The

A CASE OF OSTEOMA OF THE FRONTO-ETHMOIDAL REGION

By A. B. N. RAO (Hyderabad (Dn), India)

THIS case is reported because of the large size of the tumour and its comparatively rapid growth which impaired the movements of the eyeball and also produced nasal obstruction.

The history is as follows:

An average built young woman of 20 years was referred to the E.N.T. Department on *11.4.58* from the Ophthalmic section of the Sarojini Devi Hospital because of a swelling over the region of the left eyebrow which was presumably increasing in size during the last 6 months.

The eyeball itself was pushed forward, and its movements were restricted towards the medial side.

The other findings were as follows:

Left Eye:

Conjunctiva—Congested.
Cornea—Opacity just below the centre.
Anterior chamber—Normal.
Iris—Clear.
Pupil—Round, reacting to light.
Fundus—Normal.
Vision—6/60.

Right Eye:

No abnormality detected.

E.N.T. Examination: A hard mass was noted below the left eyebrow.

Anterior Rhinoscopy: A hard mass was noted in the left nasal cavity anterior to the middle turbinate—mucosa intact.

Post-nasal Space: Adenoid mass noted. Sense of smell not impaired. Pharynx, larynx and examination of both ears revealed no abnormality.

X-ray of the nasal sinuses showed a large opaque mass filling the nasal cavity and the fronto-ethmoidal region, extending up to the pituitary fossa. Right frontal sinuses showed a comparatively less opaque shadow (polyp?) and the left maxillary sinus also showed opacity (X-ray Figs. 1 and 2).

A provisional diagnosis of osteoma arising from the left fronto-ethmoidal region was made.

On *16.4.58* under general anæsthesia external frontal operation was undertaken.

Howarth's incision was made and the skin, muscles and periosteum were incised and retracted. The floor of the frontal sinus was absent. A hard, irregular tumour could be palpated from the outside of the œsophagus Since it appeared to be limited to the œsophageal mucosa and not to have invaded the larynx, it was decided to spare the latter The tumour was 2 cm long and involved the entire circumference of the œsophagus Three centimetres of the œsophagus was excised with a safety margin of 0 5 cm on either side of the tumour After extensive mobilization of the œsophagus end to end anastomosis was performed The subserosa was sutured with interrupted catgut stitches not passing through the mucosa The musculature was then sutured in various layers A stomach tube was passed and left *in situ* for one week The neck wound was sutured primarily, and it healed without fistulation After removal of the stomach tube the patient could swallow liquid food Owing to a tendency to narrowing at the suture line, dilatation was effected with a bougie once a week for 1 month The patient could afterwards eat solid food without trouble Now, more than 3 years after the operation, she still feels well Recent œsophagoscopy and roentgen examination still showed a slight stricture at the level of the anastomosis but no signs of a recurrence (Fig 2)

In selected cases it is thus possible radically to remove a tumour of the cervical œsophagus without resorting to laryngectomy or difficult plastic operations The method is, however, only possible if the disease is detected in good time But such early discovery is rare because as a rule œsophageal cancer does not produce symptoms until late

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FIG 1

X ray sinuses—A P view showing large opaque mass over the left fronto ethmoida region and practically filling the whole nasal cavity

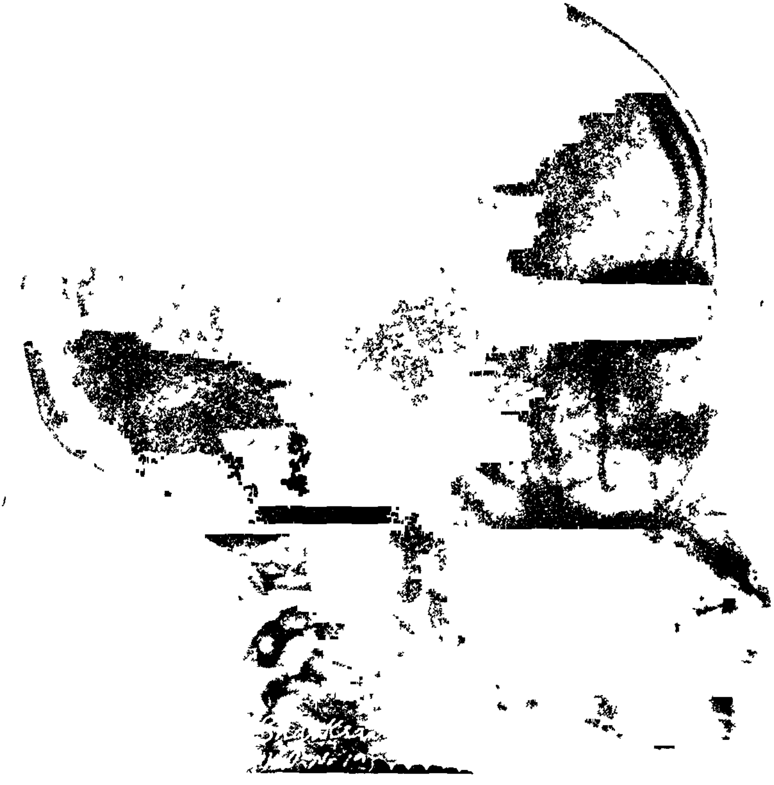

FIG 2

X-ray skull lateral view—showing the extension of the tumour up to the pituitary fossa

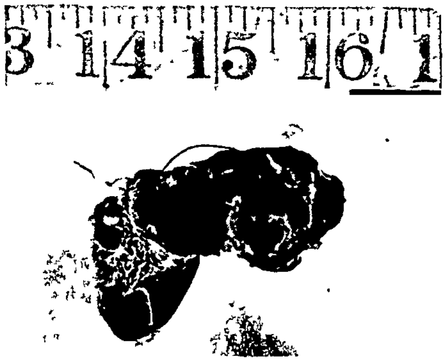

Fig 3
Photograph of tumour A P view showing lobulations

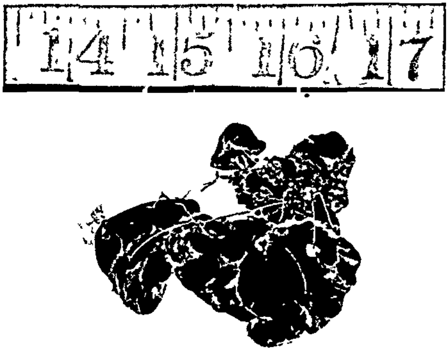

Fig 4
Photograph taken from above showing the extension backwards

tumour was noted attached to the lateral margin near the fronto-zygomatic suture. This tumour was extending through the ethmoid and protruding into the left nasal cavity. Posteriorly its extension was up to the region of the left optic foramen. Some mucoid discharge and a small polyp was also noted in the left frontal sinus. With the help of gouge and mallet the entire tumour was removed in three pieces. The left frontal, ethmoidal and sphenoidal sinuses were completely explored. The left maxillary sinus was punctured and washed. It revealed very little mucoid discharge.

The whole operation area was insufflated with penicillin powder and the wound was closed after insertion of a polythene tube in the region of the fronto-ethmoidal duct. This tube and the skin sutures were removed on the 8th day.

The patient recovered uneventfully and was discharged on the 12th day.

A follow up on *6.5.58* showed that the movements of the eyeball were completely normal and the Levator palpebrae superioris muscle was regaining its function quite satisfactorily. The nasal cavity was clear and apart from a linear scar over the region of the left eyebrow no deformity was noted.

Examination of the specimen shows that it was of very hard consistency with numerous small nodules. Microscopic appearance was that of a typical compact osteoma.

Acknowledgments

I am very grateful to Dr. P. Ramchander, Superintendent, Sarojini Devi Hospital and Institute of Ophthalmology and Otolaryngology for referring this case to me and also allowing me to publish it.

CLINICAL NOTES

NEW OPERATIVE TECHNIQUE FOR PERMANENT TRACHEOSTOMY

By ROBERT EVANS (Birmingham)

IT is difficult to believe that there can be anything new in the performance of an operation which must surely be one of the oldest surgical procedures Nevertheless when I described the following technique at a clinical meeting (Midland Institute of Otology, October, 1957), it appeared to be new to my colleagues

The usual incisions for tracheostomy are the vertical and the transverse, but there are drawbacks to both The vertical incision gives a good exposure, though the subcutaneous tissue tends to get in the way especially in fat patients It is also awkwardly placed if a laryngectomy has to be done later using a U-shaped incision With the transverse incision, exposure of the thyroid isthmus may be inadequate

To overcome these drawbacks I suggest turning down a flap of skin and subcutaneous tissue, the base of which is just above the jugular notch The transverse part of the incision should be about ¾ inch in length, slightly concave upwards and situated at the level of the lower part of the cricoid cartilage (Fig 1).

The thyroid isthmus is then exposed in the usual way, infiltrated with local anæsthetic, and divided After injection of local anæsthetic into the lumen of the trachea, a semicircular piece is excised from the anterior wall, the straight edge being between the first and second rings, and the curved edge *extending downwards to between the fourth and fifth rings (Fig 2)*

The concave margin of the skin flap is now sutured to the lower edge of the tracheal incision with five fine catgut sutures, the first being placed in the midline (previously marked on the skin) and two sutures inserted on either side of this It is easier to insert these stitches if they are not tied until all are in position

The upper part of the wound tends to spread, but the raw area can be reduced by a few well placed sutures (Fig 3)

The skin flap may appear to be somewhat bulky in some cases, but will be found to have shrunken by the following day.

Some advantages of this technique are as follows

1 It is virtually impossible for the tracheotomy tube to be displaced

2 The patient appears to be more comfortable with the tracheotomy tube resting on skin surface rather than on skin edge

3 The tube can usually be dispensed with after a week or so.

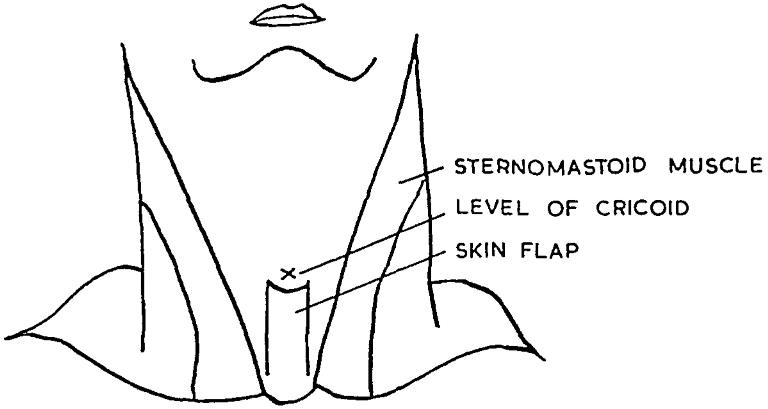

FIG. 1.
Skin Incision for Tracheostomy.

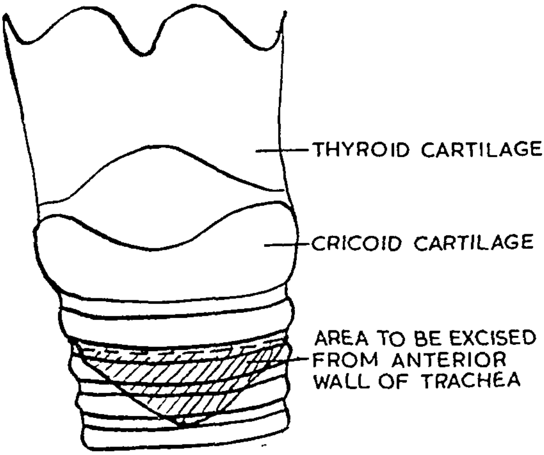

FIG. 2.
Excision of Anterior Wall of Trachea.

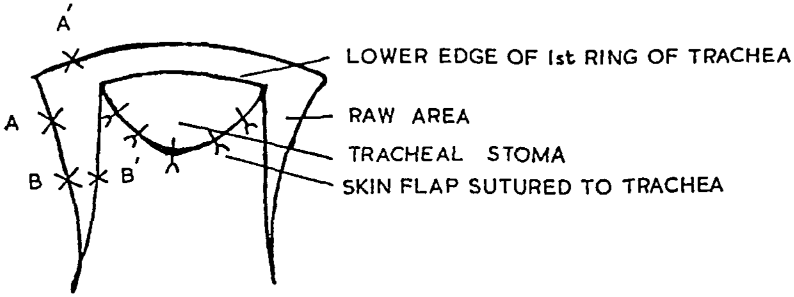
A′
A
B
B′
LOWER EDGE OF 1st RING OF TRACHEA
RAW AREA
TRACHEAL STOMA
SKIN FLAP SUTURED TO TRACHEA

FIG. 3.
Raw area reduced by suturing A to A^1 and B to B^1 on each side.

4 Epithelialization of the tract is rapid and there is less tendency to stenosis

5 The subglottic region can easily be inspected with a mirror through the large tracheal opening at the time of operation, or by means of a nasopharyngoscope inserted through the stoma subsequently.

Summary

1 A method of performing a permanent tracheostomy by using a skin flap is described

2 Certain advantages are claimed for this technique

SERO-MUCINOUS MIDDLE-EAR CATARRH : A NOTE ON TREATMENT

By B. H. COLMAN (Edinburgh)

VARIOUS forms of treatment have been employed in the management of persistent sero-mucinous catarrh of the middle ear. In the past, these have included repeated Eustachian catheterization sometimes coupled with the passage of a Weber-Liel aspirating tube or Hunter Tod gum elastic bougie if there has been any question of stenosis, in addition a variety of measures directed to the nasopharynx, nose and sinuses have been advocated. The role of allergy has been stressed, though treatment of this supposed predisposing factor is usually disappointing. Frequently, repeated paracentesis and aspiration has been necessary though not always satisfactory.

More recently, particularly in America, fine polythene catheters have been left in the middle ear usually for several weeks, sometimes even as long as six months in an endeavour to overcome the need for repeated paracentesis. The catheter can be introduced *viâ* the Eustachian tube or *viâ* a small stab incision in the drum apparently without ill-effect.

It is felt, however, that such a procedure has definite drawbacks apart from the risks of infection or physical damage of the delicate tubal or middle-ear lining (though these complications are reported to be infrequent). In particular it is considered that the fluid in the middle ear is frequently far too tenacious in consistency to be completely evacuated by these methods and in practice the following form of treatment has been found to be entirely satisfactory.

Under local anæsthesia a flap is raised as in the Rosen permeatal approach to the stapes. In all cases operated on the middle ear has been found full of glue-like material which could only be removed by the strongest suction. Clearance from the window recesses and the attic spaces can be especially difficult but it is helped by the local introduction of hyaluronidase. Eustachian inflation is carried out and not infrequently produces a further collection of the same gluey material, which is again aspirated.

Prior to replacing the drum it is convenient to leave in either hyaluronidase or cortisone to aid final resolution.

Complete return of normal hearing can be anticipated.

The following case is illustrative and of added interest on account of the apparent element of nerve deafness.

Mrs. J.M., aged 37, complained of unilateral deafness of several weeks duration which began with a cold. The drum had an opaque golden-yellow appearance, was featureless and immobile whilst investigation of the nose, nasopharynx and sinuses revealed no abnormality. Audiometric examination was carried out and in addition to the middle-ear deafness, revealed a considerable perceptive loss (As there had been slight vertigo at the onset of her disability it was therefore suggested that the initial condition had been one

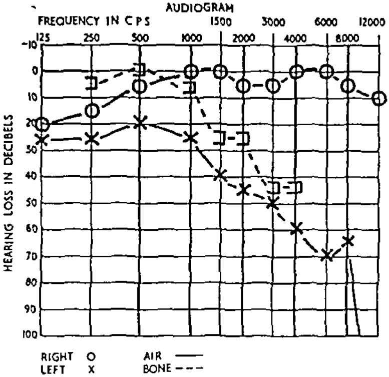

FIG I
Pre operative Audiogram

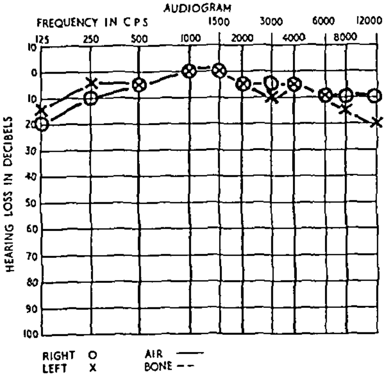

FIG 2
Post operative Audiogram

of acute otitis media with a mild serous labyrinthitis) Eustachian inflation repeated on several occasions produced a completely clear air entry but no improvement in hearing whatever, and the drum remained unchanged The condition had the appearances of an early adhesive deafness but exploration was considered worth while in view of the history. The findings at operation were similar to those just described. Pre- and post-operative audiograms are illustrated.

Return of normal hearing was immediate and permanent, there has been no further trouble in the twenty months since exploration.

Comment

In retrospect there is no doubt that in this instance the perceptive loss was the result of immobilization of the endolymph caused by the middle-ear exudate firmly blocking both windows. Such a mixed deafness is unusual, certainly in adults.

Middle-ear exploration for middle-ear deafness of uncertain cause is being increasingly practised in most clinics and is throwing much new light on the pathology of this region. It is not suggested that it is always necessary in cases where an obviously watery transudate with a movable fluid level is plainly visible but it should be borne in mind for those patients with a persistently abnormal drum and abnormal hearing whatever the condition of the Eustachian tube. Experience of over a dozen cases (in several of which Eustachian inflation gave a clear air entry) has led to a policy of early exploration rather than of waiting till other measures have been tried and have failed. The above treatment however has been found equally successful in cases of comparatively long standing, which would otherwise formerly have been candidates for a permanent and severe adhesive deafness.

SOCIETIES' PROCEEDINGS

THE SCOTTISH OTOLARYNGOLOGICAL SOCIETY

THE Seventy-Sixth Meeting of the Society was held in the Royal Northern Infirmary, Inverness, on Saturday, May 24th, 1958 The President, Dr F T Land was in the Chair

Papers were given by Dr I Simson Hall on "Otosclerosis in Fragilitas Ossium", by Dr A Brownlie Smith on "The Pathology of a Case of Traumatic Deafness in a Child", and by Mr J C Campbell on "Clinical Report on 300 cases of Chronic Suppurative Otitis Media who have undergone Endaural Mastoidectomy" A film showing fenestration, stapes mobilization and tympanoplasty was shown by Dr Finlay Brown

Sixteen cases were shown by Dr I M Seex, and three by Dr. H M Urquhart The following were of special interest

Case II *Trigeminal Neuralgia* Miss D C, aged 65, complained of lancinating pain over the left antrum She had a left facial palsy of the lower neuron type, resulting from an accident in 1942 Nose and sinuses appeared healthy There was a left sided chronic suppurative otitis media Trigeminal neuralgia affecting the second division of the Vth Nerve was diagnosed On *5 8 57*, under hypotensive anæsthesia, the maxillary division of the Vth nerve was resected from the foramen rotundum, forwards to the infra-orbital foramen, by the trans antral approach To date the patient has had no further pain

In the discussion, Dr Simson Hall recalled seeing a similar case in a girl medical student, who developed trigeminal neuralgia following an accident She was treated by posterior section, which was very successful to begin with, but the pain returned after 3 years Dr Seex said that he had done a number of these cases over the past 7 years by the trans-antral route, and did not experience the return of pain One case, labelled as a neurotic had been operated on five years ago by dissection of the spheno palatine ganglion, and had been free from pain since then

Case III *Pachydermia of the Vocal Cords* A woman, aged 26 was first seen in *February, 1958*, complaining of loss of voice of six weeks' duration Both vocal cords appeared thickened and covered with inflammatory exudate X-ray of chest revealed no disease Wassermann and Kahn reactions were negative On direct laryngoscopy, the cords appeared irregular and ulcerated Biopsy report stated "inflamed mucosa covered with hyperplastic squamous epithelium This showed irregular thickening with elongation and fusion of rete processes Moderately active mitosis was present The surface was covered with necrotic keratinized debris and loosened cells, infiltrated with polymorphs There was chronic inflammation of the deeper parts There was no evidence of tumour, nor of specific type of inflammatory lesion". The patient was 8-months pregnant, and her mother suffered from a similar hoarseness during her pregnancy.

Dr. Seex thought that the diagnosis rested between a pseudo-malignant hyperplasia, an inflamed pachydermia or a pre-malignant lesion.

Dr. Simson Hall considered this to be an endocrine change. He had seen several such cases in the Simpson Memorial Hospital, Edinburgh. The hoarseness cleared up after the pregnancy.

Dr. Finlay Brown thought that the condition might be allergic, similar to the allergic rhinitis which often occurred in pregnancy.

Case XV. *Trans-antral Ethmoidectomy.* A man, aged 76, was first seen in *November, 1953,* with bilateral nasal polypi, allergic nasal mucosa, and opacity of antra and ethmoids to X-ray. There was a history of severe asthma of several years' duration. A resection of the septum was performed, together with bilateral radical antrostomy and trans-antral ethmoidectomy. This was followed by a course of nasal ionization. No further asthma to date has been noted.

Mr. Weir congratulated Dr. Seex, and asked what his indications were for such widespread surgery. *Dr. Seex,* in reply, said that most cases of asthma were primarily due to nasal allergy. Most of them have thickened or polypoid mucosa, or even associated sepsis. It is necessary to do meticulous surgery, clear out the antra completely, and then give ionization to reduce the allergy. He had treated 100 consecutive cases on these lines, with a large percentage of cures. In the past, results had not been obtained due to X-ray films being wrongly reported as clear. In the past 16 years he had come to the conclusion that the more radical the treatment, the better the chances of cure.

OBITUARY

DR CHEVALIER JACKSON

IT is fitting that an obituary notice of Dr Chevalier Jackson, who died on 16th August, at the advanced age of ninety-two, should take the form of personal memories, rather than that of a formal curriculum vitae

In 1923 Sir StClair Thomson urged me to visit Philadelphia and to apply for a place in one of the courses of instruction given there, he impressed on me the necessity of avoiding delay, for the reason that Dr Jackson's health was somewhat fragile and that he might consequently have to abandon his teaching activities I had just previously met Dr Gabriel Tucker, his warm hearted and energetic associate, who told me of Dr Jackson's early struggles in Pittsburg, where he attempted to establish as a speciality the art and science of Bronchoscopy and Œsophagoscopy, apparently under financial embarrassment

As a young and ignorant clinical assistant I was successful in obtaining a place in his course in the summer of 1923 my reception was most cordial and Dr Jackson had gone so far as to take a room for me at one of the clubs and to instruct them to send the account to him, an act of generosity which I was naturally unable to accept, but which impressed me deeply

I was not the first from England to visit him, for as far back as 1913 Mr Howarth had paid a visit to him at Pittsburgh

The experiences gained in this Summer Course have served me to the greatest advantage throughout my medical life and very many of my friends and colleagues in the British Isles owe a similar debt of gratitude to this great man

Fragile as he was in physique, yet his energy was enormous, and even in the two days of each week which he spent in bed, to conserve his strength, were occupied in the practice of endoscopic procedures, as well as in writing

At that time the removal of foreign bodies from the air and food passages owed its elaboration to the investigations of laryngologists, among whom Jackson was one of the early pioneers, as I said at the time, in writing a short account of the course, hours and even days were spent in preparation and planning and minutes or even seconds in the removal of foreign bodies, so skilful was his wizardry, as MacKenty of New York described it

But of greater benefit to medicine as a whole were his researches and descriptions of the physiology and pathology of the air passages, of the lungs, and of the œsophagus, direct examination enabled him to interpret the mechanisms in question and to explain the opening and closure of the bronchi, the reasons for physical signs and also the control of the mouth of the œsophagus

Chevalier Jackson was born at Pittsburgh in 1865 and qualified as M D at Jefferson Medical College, Philadelphia in 1886, at the early age of 21, he practised medicine in Pittsburgh and was elected Professor of Laryngology there in 1912; the struggle must have been hard and in 1916 he returned to his Alma Mater, where he became Professor of Laryngology. Subsequently, in 1924, a chair of Bronchoscopy and Esophagoscopy was created for him. He was also professor of Bronchoscopy and Esophagoscopy in the University of Pennsylvania, a post especially instituted for him; I well remember the rush from one hospital to the other with a rapid lunch in a Cafeteria in the company of Gabriel Tucker and the genial Louis Clerf.

As many know a special clinic was later established in Temple University, where his son well-known to many as C.L., reigns today.

Chevalier Jackson was a founder of the America College of Surgeons, and in this he showed the breadth of his outlook, as in many other directions connected with the wider aspects of Medicine and Surgery.

He was Officer of the Legion of Honour and Laureate of the Academie de Medicine of Paris, in addition to innumerable other distinctions; of especial interest to us were his election as honorary Fellow of the Royal Society of Medicine of London, honorary member of the Scottish Society of Otology and Laryngology and Honorary Fellow of the Royal College of Surgeons of Edinburgh, an honour conveyed to him by his son in his absence, at the time of the International Congress of 1949.

A distinction which must have given him pleasure was the receipt of the Medallion of the American College of Chest Physicians in 1952, in recognition of his services to general medicine; he must also have been gratified to be appointed as Doctor of Humane Letters at Jefferson Medical College.

So passes a great man and so ends an epoch; thoracic medicine and surgery have advanced to an extraordinary degree and the art and science of these branches is wide-spread beyond the field of the laryngologist and early endoscopist. No greater memorial could remain than the recognition by the Medical profession in many countries of the immeasurable possibilities unfolded by this pioneer and by the untold benefits resulting from his teaching and example.

V. E. NEGUS.

GENERAL NOTES

The Editors are anxious to improve the clinical aspect of the Journal and would welcome more reports of interesting cases These clinical records need not be at all elaborate and it would be unnecessary to go minutely into the ætiology or the literature of the subject Senior Registrars have unique opportunities for seeing a large variety of cases, many with unusual features It is in many ways regrettable that these should go unrecorded It is hoped that all those in charge of clinics will help us in this matter and see that such cases are not overlooked It is important that all illustrations should have captions and also that the author's name should be written on the back

ROYAL COLLEGE OF SURGEONS OF ENGLAND
OTOLARYNGOLOGY LECTURES 1958–59

The following lectures, arranged jointly by the Royal College of Surgeons of England and the Institute of Laryngology and Otology, will be delivered in the Lecture Hall of the College, at 5 30 p m

1958
December 4th Sir Stanford Cade "Cancer of the tongue "

1959
February 5th Mr Victor Riddell "Thyroid gland and recurrent laryngeal nerve "
March 5th Mr Robert Brain "Œsophageal replacement '
April 30th Professor Martin Rushton "Cystic conditions of the upper jaw "
July 2nd Dr Denis Hill "The value of electro encephalography in diagnosis "

INTERNATIONAL SYMPOSIUM

Milan, December 7th and 8th, 1958

International Symposium on the Modern Trends in Diagnostic Radiology in Oto-rhino-laryngology

This meeting will be held under the joint presidency of Professor Luigi Pietrantoni, O R L Clinic, Milan University and Professor Arduino Ratti, Institute of Radiology, Milan University

For information about this meeting write to
Dr Carlo Agazzi or Dr Luigi Ferri, Clinica Otorinolaringologica dell'Università di Milano, Via Commenda 16, Milano, Italy

General Notes

THE INSTITUTE OF LARYNGOLOGY AND OTOLOGY

330-332 Gray's Inn Road, London, W.C.1.

The Academic Board announces the following

SPECIAL LECTURES AND DEMONSTRATIONS

to be given at the Institute—except where otherwise stated. These are open to all members of the Specialty and to postgraduate students. No ticket of admission is required.

Saturday, 8th November, 1958, at 11.30 a.m.
Professor Lucius Rüedi (Zurich)—"New developments in the surgery of otosclerotic disease".

Friday, 14th November, 1958, at 4.30 p.m.
Mr. T. G. Wilson (Dublin)—ANNUAL ADDRESS—"The modern treatment of deafness in historical perspective".

Saturday, 6th December, 1958, at 11.30 a.m.
Dr. J. A. Sullivan (Toronto)—"Modern temporal bone surgery".

Friday, 12th December, 1958, at 5.30 p.m.
Professor W. St. C. Symmers (Charing Cross Hospital Medical School)—"Mycotic diseases".

Provisional List of Short Courses for Consultants and Senior Registrars

17th, 18th and 19th February, 1959—Malignant diseases of the ear, nose and throat.

20th and 21st February, 1959—Pathology of the ear, nose and throat.

10th, 11th and 12th March, 1959—Diagnosis and treatment of the deaf child.

13th and 14th March, 1959—Modern methods of acoustic and vestibular testing.

10th and 11th April, 1959—Practical Aural Surgery Class.

The number of entries is limited in each course.

A detailed syllabus of any of the above can be obtained on application to the Secretary.

General Notes

SEMON LECTURE

THIS lecture will be delivered by Professor Lucius Ruedi of Zurich in the Barnes Hall of the Royal Society of Medicine on Thursday, 6th November. Subject Some observations on the Histology and Functions of the Larynx.

EXAMINING BOARD IN ENGLAND
BY THE
ROYAL COLLEGE OF PHYSICIANS OF LONDON
AND THE
ROYAL COLLEGE OF SURGEONS OF ENGLAND

DIPLOMA IN LARYNGOLOGY AND OTOLOGY

THE Secretary reports to the two Royal Colleges that at the First Part of the Examination held in June, 1958, 47 Candidates presented themselves, of whom 26 passed, and that at the Second Part of the Examination held in June/July, 1958, 40 Candidates presented themselves, of whom 22 passed

The following 22 Candidates, having complied with the regulations of the Board, are eligible to receive the Diploma, viz

Name	Qualification	Address
Aggrawal, Lajpat Rai	M B , B S Punjab	c/o Institute of Laryngology and Otology, Gray's Inn Road, W C 1
Alam, Badiul	M B , B S Dacca	62, Berners Street, Ipswich, Suffolk
Anderson, Arthur Alfred	M B , B S Ceylon	53, Comeragh Road, W 14
Batchelor, Peter John	M B , B S London (Middlesex)	Princess Mary's R A F Hospital, Halton, Bucks
Bloomer, Peter George Appleyard	M B B Ch Cambridge (Westminster)	2, Lupus Street, S W 1
Bloomfield, Robert Carnwath	M B , B Ch Belfast	64, Bristow Park, Belfast
Buffin, John Terence	B M , B Ch Oxford	5 Rose Crescent, Richmond, Yorks
Butler, Richard Michael Curtis	M B , B S London (Guy's)	Derwent House Whatstandwell, Matlock, Derbyshire
Chandrasekhar, Shroff Rama Setty	M B , B S Madras	c/o Mr S R Vasudev, Fraserpet Coorg India
Choa, George Wing Sien	M B , B S Hong Kong	Queen Mary Hospital, Hong Kong
Cox, Michael Gordon	M B , B Ch Cambridge L R C P , F R C S (St Thomas's)	102, Lexham Gardens W 8
Crowe, Timothy Joseph	M B , B Ch , N U I (Dublin)	Box 869, Ndola, N Rhodesia
Gabriel, Cyril Edward	M B , Ch B Glasgow	98, Otago Street, Glasgow, W 2
Ghosh, Dwijendra Prosad	M B , B S Calcutta	211, Rufford Road, Crossens, Southport Lancs
Godfrey, David John	M B , Ch B Glasgow	2, Kingswood Road, Shortlands, Near Bromley, Kent

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The Journal of Laryngology and Otology

(*Founded in* 1887 *by* MORELL MACKENZIE *and* NORRIS WOLFENDEN)

November 1958

MALIGNANT GRANULOMAS OF THE NOSE AND PARANASAL SINUSES

By C PETER MILLS (London)

PART I

Historical Survey

AT *a meeting of the Laryngological Society of London, held* on December 9th, 1896 Dr P McBride presented "a case of rapid destruction of the nose and face" The patient was a 20 year-old house painter who developed an ulcer inside his left nostril one week after scratching it When he died 18 months later destruction of his face was extensive bone being exposed from malar to malar, the whole external nose and most of the upper lip were destroyed

Bacteriological examinations, inoculations for tuberculosis and glanders left the diagnosis in doubt Many local and systemic treatments in vogue at the time were tried, but failed to stem the destructive lesion

Dr Muir, pathologist to the Royal Infirmary, Edinburgh, examined the slides and was of the opinion that the disease did not correspond to any with which he was familiar

Twenty five years elapsed before Sir Robert Woods (1921) wrote his "observations of malignant granulomas of the nose' He recorded that the first of his two cases was "as puzzling to the pathologists as to the clinicians It seemed as though a wave of granulation tissue advanced irregularly into healthy parts breaking down behind as it advanced in front, so there was never any great depth of pathological growth present"

Arrowsmith (1921) and M'Arthur (1925) reported single cases similar to those described by Sir Robert Woods.

Chattelier (1929) gave the name Lupus Pernio to a very similar condition.

Many eminent pathologists in America studied the sections of "a case of mutilating granuloma of the nose and face with fatal ending", presented by George B. Wood (1931). Dr. Eugene A. Case believed that generically it should be placed amongst the sarcomas. Ewing said that "unless we have entirely new diseases we are thrown back on mycosis fungoides or syphilis"; and Dr. F. de F. Weidman thought that it probably belonged to the so-called granulomas.

Klinger (1931) was the first to describe the condition of rapid destruction of the nose and ethmoids associated with diffuse lesions of periarteritis nodosa.

Four more cases of localized malignant granuloma reported by Stewart (1933) were used in a very comprehensive paper on the condition up to that date. He elucidated the salient points in the clinical picture where the primary disease was in the nose.

Berendes (1934) thought the condition was on the increase in Germany, already 15 had been recorded. He quoted Kraus (1929), Hennig and Wirth (1926) as describing similar cases, but on examination of these papers they do not conform to the disease of malignant granuloma as it is now known.

Simson Hall (1934), Bayer (1937) and Schütz (1938) are often cited as having described further cases; however, they cannot be included in this series because of their unusual features which do not fit into the general pattern.

Joisten (1936), Lewy (1938) and Heinermann (1938) described a further four cases in which the disease originated in the nose.

In 1939, Wegener reported three cases of what is now known as Wegener's granulomatosis. They all developed a peculiar destructive condition of the nose, and at death were found to have widespread periarteritis nodosa. Apart from Klinger's 1931 report, this was the first major paper in which the conditions of malignant granuloma and periarteritis nodosa were associated.

In a paper by Hoover (1941) recording a further 5 cases of the localized disease he noted that the common organisms found in the granulomas were staphylococci, streptococci and diphtheroids.

Lierle (1942) and Isbell (1943) reported a further 2 cases.

In 1945 Hargrove *et al.* said it was impossible to decide whether the condition was due to an unidentified infection or to an unusual type of sarcoma change. In the same year Rasmussen remarked that it is indeed a striking feature that in a previously healthy person such a violent and destructive process may develop without eliciting any reactive phenomena on the part of the organism.

Malignant Granulomas of Nose and Paranasal Sinuses

H L Williams read a paper before a meeting of the American Laryngological Association in 1949 on "lethal granulomatous ulceration involving the midline facial tissues', and surveyed the whole literature on the subject He described 3 of his own cases, all of which died despite tremendous efforts to cure them effectively

Bergvist (1949) recorded 2 more cases, and Williams *et al* (1950) reported one in which a biopsy at death revealed a squamous epithelioma (personal communication)

Howells and Friedmann (1950) in a paper primarily discussing the pathology of giant cell granuloma and periarteritis produced evidence that the nose or paranasal sinuses can be the first site of the peculiar granulomatous reaction In a later paper Friedmann (1955) discussed more fully the pathology of malignant granulomas of the nose

McCart and Woodburn (1950) reported 2 and 6 cases respectively, three of which were associated with periarteritis nodosa

On the advent of cortisone and A C T H more papers were published advocating the use of these drugs in the treatment of malignant granuloma Moore's (1951) case was alive 18 months after treatment with cortisone, this patient has since died (personal communication)

A case treated with corticotropin for 25 days by Hagens *et al* (1953) was still alive and free from disease in 1956 (personal communication)

Alexander (1954) and Breckenridge in the same year were unsuccessful in treating their case with A C T H and cortisone

Further additions to the literature on the Wegener s granulomatous type have been made by Stratton *et al* (1954) 2 cases, Fahey *et al* (1954) 4 cases, Seidelin *et al* (1954) 2 cases McCallum (1954) 1 case, and Geist (1953) 1 case

The cure of a patient suffering from a granulomatous ulcer of the nose when treated with chloramphenicol was reported by Sneddon *et al* (1952) This patient died 18 months later with a recurrence in the pharynx (personal communication)

Levan (1953) added 2 more cases and reviewed the theories on the ætiology He quoted Williams who mentioned the possible relationship to the so called collagen diseases and Woodburn and Harris who thought that the nasal condition was a localized manifestation of a systemic periarteritis nodosa

Paterson (1956) was unable to add any new evidence either with regard to ætiology or treatment when he reported 6 more cases

Maxwell Ellis (1956) has been able to record a case that had extensive destruction of the nose by malignant granuloma in 1947, and who is now quite well and free from the disease after small doses of radiation

That radiation may be the treatment of choice in the future is borne out by Gilroy Glass (1955), 3 cases, and Howells (1955) whose 2 cases have apparently been cured by small doses of X-rays

The Clinical Picture

Age and Sex Incidence

The small number of cases recorded in the literature make it impossible to obtain figures that are statistically significant. Where authors have given any indication as to age and sex incidence, they have all disagreed.

Of the 86 cases reviewed in this series, 55 were males, and 31 females, There is enough disparity between these figures to indicate that the disease affects males in a far greater proportion than females.

The age incidence does not vary in the two sexes, the majority of cases being found between 25 and 55 years, the greatest number occurring in the late thirties and early forties (Fig. 1).

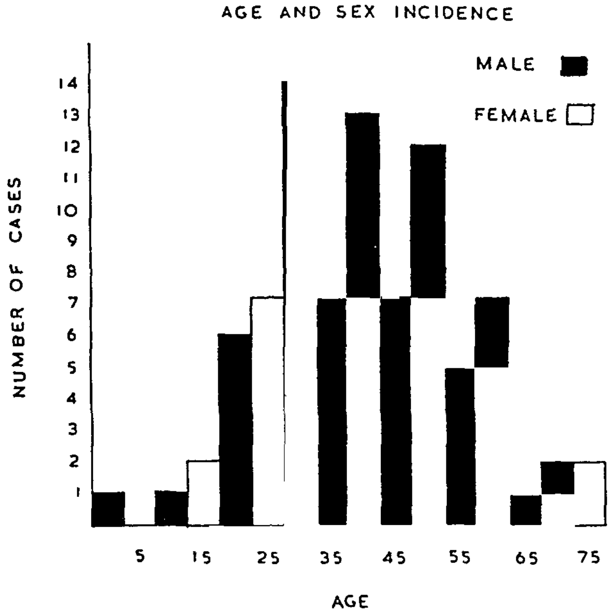

FIG. 1.

Age and sex incidence in 86 cases of malignant granuloma.

Primary Site

This survey deals with cases where the nose or paranasal sinuses have been involved as the primary site. The disease was judged to start in the nose of 67 (77·8 per cent.) patients, and in only 19 (22·2 per cent.) did it begin in the antrum or very rarely, one of the other paranasal sinuses.

Previous History

A careful examination of the previous medical histories of all these cases repeatedly reveals a preceding paranasal infection of long standing, in one case, twelve years These infections have given rise to a stuffiness of the nose, nasal obstruction with, or without accompanying watery discharge, to such an extent that relief was sought through surgical means Submucous septal resection, with or without drainage of the antrum, has been performed in a substantial group of these patients

Stewart prefers to call this the prodromal stage, in which no active disease is present

Non-surgical trauma has been suggested as a starting point in some cases, but this was recorded so infrequently, and its occurrence in normal life so common, that it must be excluded as a contributory factor

That the disease became apparent in many patients soon after nasal operations to relieve obstruction, makes it appear probable that the condition was already present at the time of the surgical intervention, but was not recognized

Symptoms

Where describing the symptoms, it is simpler to divide the disease into three main groups

(1) Where the nose is the primary site
(2) Where the antrum is the primary site
(3) In cases of periarteritis nodosa where there is an associated nasal or paranasal granulomatous lesion

Because of the close anatomical relationships there are many symptoms common to each group

Nose In practically every case stuffiness or blockage of the nasal airway was complained of A watery or serosanguinous discharge, which may or may not be blood-stained is a usual accompanying feature On a *few occasions an epistaxis is the main symptom* Thereafter the symptoms depend entirely on the stage of the disease, and the anatomical site involved

Swelling of one side of the nose, or at the inner canthus of the eye, either painful or painless, or a sore in the vestibule which gradually spreads in spite of treatment, are often associated with a nasal obstruction, serosanguinous discharge or an epistaxis If a secondary infection has occurred, the lips, face, and periorbital tissues may be grossly swollen and painful

Paranasal sinuses Not infrequently these patients have either been referred from, or to a dental surgeon because of persistent maxillary *toothache After the underlying antral disease has been discovered when* dental extractions have failed to alleviate the toothache, swelling over the cheek, and pain, local or referred to the ears are usual, as are frontal headaches Symptoms referrable to the nose are common, simulating those of sinusitis because of the super-added infection Once the disease has spread to involve the nasal cavities frequent small epistaxes and a watery discharge are complained of. When the floor of the orbit has been invaded, a diplopia, and neuralgia or paresis of the infra-orbital nerve may occur. Trismus secondary to involvement of the muscles, or even direct invasion of the temporo-mandibular joint has been recorded. This was a very prominent symptom in two of my cases. The persistent post-nasal discharge causes anorexia or vomiting.

Periarteritis nodosa: Geist and Wylie reported a case, whose main complaint for two months had been of multiple smooth, dull red papillae scattered over the trunk and buttocks. A granuloma was found in the nose on clinical examination. A history of nasal congestion and rhinorrhœa for two years was elicited. At autopsy an acute necrotizing arteritis was revealed, involving kidneys, skeletal muscles, lungs, œsophagus, and small intestine, in addition to the non-specific chronic inflammatory process of the upper respiratory passages.

McCallum's patient felt unwell, with chills, shivering and sweating attacks, and had vomited on two or three occasions. A week later he noticed a slight swelling and tenderness of the cheek and nose. This patient died, and was found to have periarteritis nodosa, associated with a granuloma of the nose.

Signs

The physical findings vary, depending on the position and stage of the disease. The noticeable clinical feature that many authors have commented on, is the apparent well-being of the patient, despite in some cases, advanced disease, with gross disfigurement. It is quite remarkable how little constitutional disturbance there is, more especially when one considers the lack of the usual signs of resistance on the part of the body, as shown by fever, anorexia, loss of weight, or even leucocytosis.

Where the disease is involving the nose, the mucosa is swollen and covered by glutinous crusts, on removal of which, masses of what appears to be simple granulation tissue are revealed. There may even be exposure of bone. Associated with these findings, a peculiar odour has frequently been recorded. As the condition progresses, ulcerations develop on the chonchae and septum. Sequestra form and have to be removed. The hard palate is often invaded, and large, rapidly spreading ulcers supervene. A secondary infection of the soft tissues causes œdema of the lips, cheeks, and eyelids; and abscesses may develop. Massive erosions of the nose cheeks, lips and hard palate soon follow. This mutilating destruction was extreme in Chattelier's case, and almost as great in McBride's. In these cases "the hard and soft palates were destroyed and through the mouth, the roof of the nose and nasopharynx were visible. The roof of the maxillary sinus was exposed by destruction of the lateral wall of the nose. The alveolar processes were ulcerated, and the teeth either dropped out or were loose". As Stewart remarked, the facial appearance is monstrous.

Numerous small hæmorrhages are frequent, if larger vessels become involved a massive hæmorrhage may terminate fatally With the advent of secondary infection and the difficulty of getting adequate nutrition cachexia is marked, the patient dying of exhaustion or bronchopneumonia When the meninges have been invaded a meningitis has been the cause of death

Where the antrum is the primary site of the disease, localized tenderness over the cheek, as in acute sinusitis may be the first sign As the disease advances, ulcerating perforations into the nose and alveolus are common The pterygo maxillary and temporal fossae are invaded trismus develops When the posterior and lateral walls of the antrum have been destroyed pains referred to the temporal and zygomatic regions are complained of Once the disease has broken the bony confines of the antral wall, the progress is very similar to that in the nose

X-ray Findings

In a paper on "The Roentgenologic Aspects of Lethal Granulomatous Ulceration of the Midline Facial Tissues ', Geist and Mullen, summarized by saying that the lethal granuloma cannot be distinguished from a neoplasm or specific granuloma on the basis of Roentgenograms

Where bone erosion has been observed prior to a biopsy of the granuloma, a malignant neoplasm has always been diagnosed

X rays are invaluable when judging the extent of the disease, after a diagnosis has been made, to facilitate more accurate treatment

Routine chest X-rays should be done They may reveal the first signs of a generalized periarteritis nodosa involving the lungs

Case Reports

CASE I, a female, aged 29 years

November 1951 She complained of a sudden pain in and behind the right ear, and above the right eye of two weeks' duration An examination revealed no abnormality apart from a carious right upper molar tooth The tooth was extracted but there was no alleviation of the symptoms after one month The pain had spread into the occipital, frontal and right facial regions A mild trismus was present An X-ray showed the right antrum to be opaque A proof puncture of this antrum revealed a fair amount of thick, blood stained, odourless pus which on culture grew a few colonies of *staphylococcus aureus*

The serum and C S F Wassermann reactions were negative, there was a mild iron deficiency anæmia A chest X ray was normal

A course of 300,000 units of distaquaine penicillin daily was started

December 5th, 1951 Under general anæsthesia the right autrum was explored The mucosa was noted to be thickened and friable, it looked malignant. There was an erosion of the postero-lateral wall of the antrum, exposing the temporalis muscle.

Histologically the material was found to be entirely fibrous connective tissue devoid of any epithelial lining, but covered in places by a necrotic slough. There was a very heavy chronic inflammatory infiltration.

January, 1952. A further X-ray of the sinuses showed the postero-lateral wall of the right antrum to be absent. The antrum was re-explored and found to be full of a pale friable "growth". The erosion previously noted had become larger. The condition was arising within the antrum and spreading outwards.

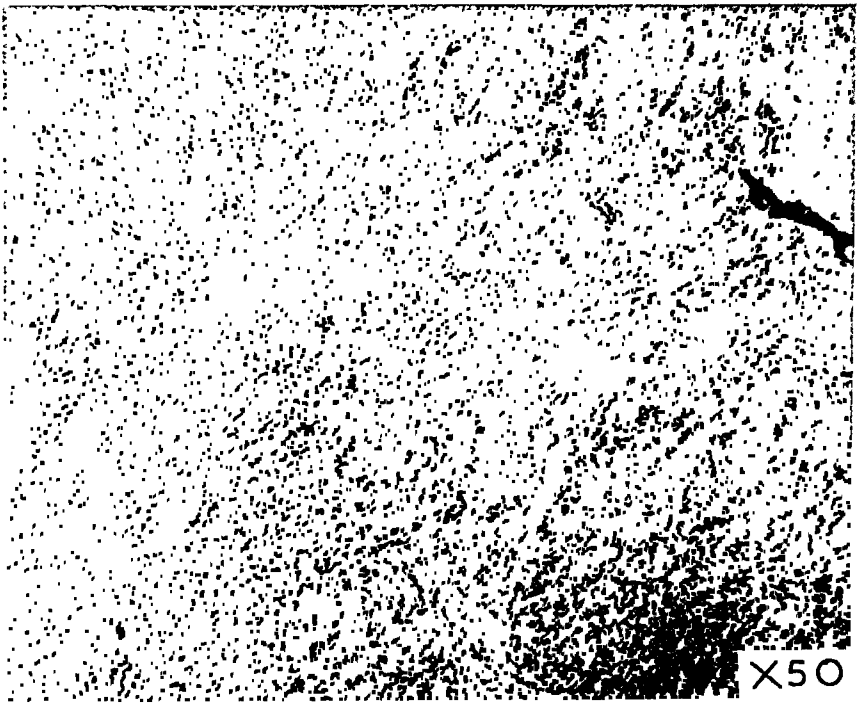

FIG. 2.

A section from the "growth" showing heavy infiltration by leucocytes into the densely collagenous material. × 50.

Sections taken on this occasion consisted mainly of granulation tissue, richly infiltrated by polymorphs and leucocytes. In those areas that bordered the antral wall, the tissue was fibrillary, whorled and densely collagenous (Fig. 2).

February 1st. In spite of the negative histological findings the case was treated as a carcinoma of the antrum, by high voltage deep therapy.

Over a period of 14 days a tumour dose of 2,619 "r" was given There was some improvement in her general condition within seven days

April 8th There was no macroscopical evidence of a recurrence when examined under general anæsthesia A biopsy showed the appearances of typical radiation fibrosis

May 20th Because of a recurrence of pain and swelling in the right cheek a further examination was made the inferior turbinate was removed, sections of which showed the same puzzling pathology as before the pathologist reported that similar conditions had been classified as malignant granulomas

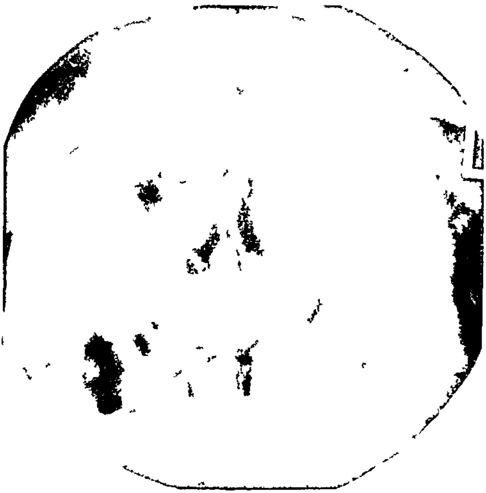

FIG 3
Shows gross destruction of the lateral wall of the antrum

In view of a paper by Sneddon (1952) on the treatment with success of this condition by chloramphenicol, the patient was given a long course of the drug totalling 44,000 mg

November 11th More biopsies showed the disease to be unchanged A further course of chloramphenicol was instituted

April 1953 The patient was readmitted with a worsening of the symptoms The right eye had become proptosed, there had been a transient diplopia and the bridge of the nose had fallen in There was much crusting in the right antrum. An X-ray showed gross destruction of the lateral wall of the antrum (Fig. 3).

Within one month a generalized abdominal pain developed, associated with a tender palpable mass in the right iliac fossa. This was thought to be neoplastic rather than inflammatory.

July. Daily injections of 5 c.c. of A.C.T.H. were given for 5 days, 4 c.c. for 10 days, and 3 c.c. for 3 days. On the second day of the course a purpuric rash appeared on all limbs, within 3 days it had faded and the mass in the right iliac had disappeared. The antral condition remained unchanged.

July 26th. A severe epistaxis necessitating a blood transfusion occurred. The patient died the following day. Permission for autopsy was not given.

Investigations

Blood. Many hæmoglobin and white blood cell counts were performed. Apart from a mild iron deficiency anæmia no abnormality was detected. The eosinophils reached 3 per cent. on one occasion. E.S.R.s estimated during the last 6 months of life varied between 50 and 69 mm. fall in the first hour. The blood urea was always within normal limits.

Urine. Routine weekly urinalyses were normal.

Bacteriology. Innumerable swabs were taken from the nose and antrum, culture of which invariably grew a *staphylococcus aureus,* sensitive to all antibiotics.

CASE II, a male, aged 16 years.

May 1954, he was admitted to the Royal Berkshire Hospital, Reading, with a pyrexia of unknown origin.

Previous history. 1944 his tonsils and adenoids were removed. 1952 he had a bilateral excision of cervical tuberculous glands.

Investigations. Because of the previous tuberculous history an E.S.R. estimation was carried out and found to be 126 mm. fall in the first hour. The blood picture was normal. No abnormality was detected in the urine. A culture from a gastric lavage grew no acid-fast bacilli. X-rays showed the chest to be normal, but the right antrum was opaque. A proof puncture of the antrum revealed a blocked ostium. Penicillin and 1 per cent. ephedrine were instilled through an indwelling polythene catheter. The fever subsided, but toothache in the right upper jaw was complained of. Further X-rays and tomograms now showed the right antrum to be opaque, with an erosion in the outer wall.

The antrum was explored and found to be full of "growth". The outer antral wall was eroded. The histological report said it might be interpreted as a chronic inflammatory reaction, or if it were a tumour, a plasma cell myeloma. After completing a 10-day course of penicillin the patient returned to his home in Bristol.

May 12th. There was a tender diffuse swelling over the right maxilla. The right inferior turbinate was enlarged and congested. The soft palate was retracted on the right side. An X-ray showed no change from that previously reported (Fig. 4).

July 14th. Numbness in the right cheek, upper lip and nostril, together with a horizontal diplopia, had developed a few days previously. His sleep had been disturbed by a nagging pain in the right temporal, maxillary and occipital regions. Pain on the right side of the hard palate caused dysphagia. A profuse foul-smelling post-nasal discharge was giving rise to anorexia and nausea.

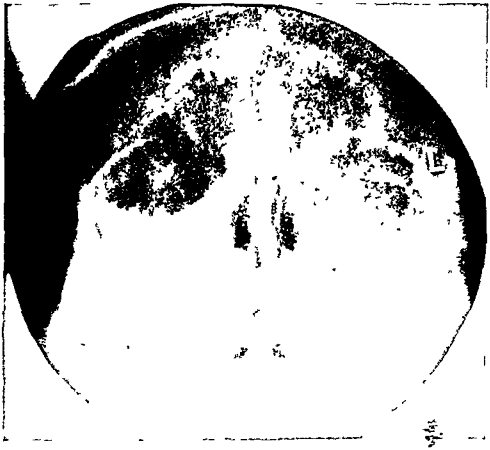

Fig 4

Shows destruction of the roof, lateral and medial walls of the right antrum

He looked pale and unhealthy, with a slight malar flush. There was a fullness over the right maxilla, and the right eye was pushed upwards (Fig. 5). Trismus was marked due to pain and fixation of the right temporo-mandibular joint. The teeth looked healthy but were tender to touch in the right upper jaw. The soft palate on the right was retracted and fixed. There was some pus in the post-nasal space. The right inferior turbinate was congested; no granulations or pus were seen on anterior rhinoscopy.

crusting in the right antrum. An X-ray showed gross destruction of the lateral wall of the antrum (Fig. 3).

Within one month a generalized abdominal pain developed, associated with a tender palpable mass in the right iliac fossa. This was thought to be neoplastic rather than inflammatory.

July. Daily injections of 5 c.c. of A.C.T.H. were given for 5 days, 4 c.c. for 10 days, and 3 c.c. for 3 days. On the second day of the course a purpuric rash appeared on all limbs, within 3 days it had faded and the mass in the right iliac had disappeared. The antral condition remained unchanged.

July 26th. A severe epistaxis necessitating a blood transfusion occurred. The patient died the following day. Permission for autopsy was not given.

Investigations

Blood. Many hæmoglobin and white blood cell counts were performed. Apart from a mild iron deficiency anæmia no abnormality was detected. The eosinophils reached 3 per cent. on one occasion. E.S.R.s estimated during the last 6 months of life varied between 50 and 69 mm. fall in the first hour. The blood urea was always within normal limits.

Urine. Routine weekly urinalyses were normal.

Bacteriology. Innumerable swabs were taken from the nose and antrum, culture of which invariably grew a *staphylococcus aureus*, sensitive to all antibiotics.

CASE II, a male, aged 16 years.

May 1954, he was admitted to the Royal Berkshire Hospital, Reading, with a pyrexia of unknown origin.

Previous history. 1944 his tonsils and adenoids were removed. 1952 he had a bilateral excision of cervical tuberculous glands.

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The antrum was explored and found to be full of "growth". The outer antral wall was eroded. The histological report said it might be interpreted as a chronic inflammatory reaction, or if it were a tumour, predominated though unlike a genuine plasma cell myeloma In the deeper areas bulky fibrous tissue trabeculae were interspersed by cellular fibroblastic elements which were relatively free from inflammatory cells (Fig 6)

On the 13th post-operative day a course of high voltage therapy was started to the right antrum and orbit A total tumour dose of 4 553 r was given in 22 treatments over 30 days Two months after the operation he was discharged

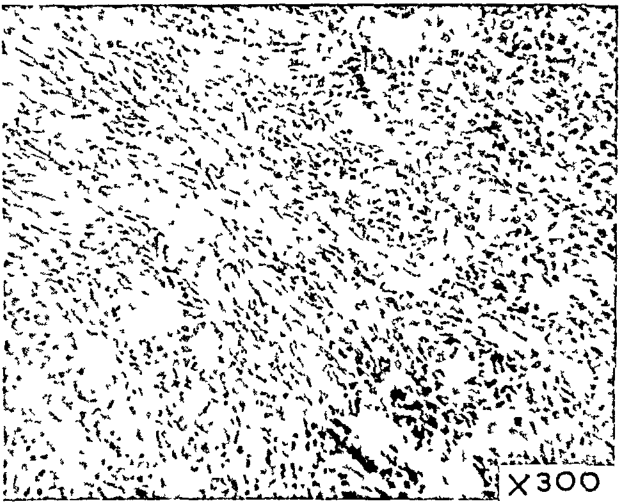


FIG 6

Section of the granulations showing the fibro cellular stroma × 300

November 22nd A biopsy was taken from the edge of the oro antral fistula and in view of the possible relationship between this disease and periarteritis nodosa a biopsy was taken from the right pectoralis major muscle No abnormality was detected in either of these two biopsies

The patient was last seen in March 1958, he was quite fit and there was no evidence of a recurrence

Investigations

Blood Throughout the time in hospital weekly blood examinations were carried out, later monthly, and finally six monthly The total and differential white blood cell counts, and the hæmoglobin were always within normal limits; there was never an excess of eosinophils. The E.S.R. is recorded graphically in Fig. 7.

Pus from the antrum grew a *staphylococcus aureus* on numerous occasions.

Urine. Frequent urinalyses were performed and showed the specimen to be acid, but no other abnormality.

X-rays. Routine chest X-rays were carried out, no abnormality was detected.

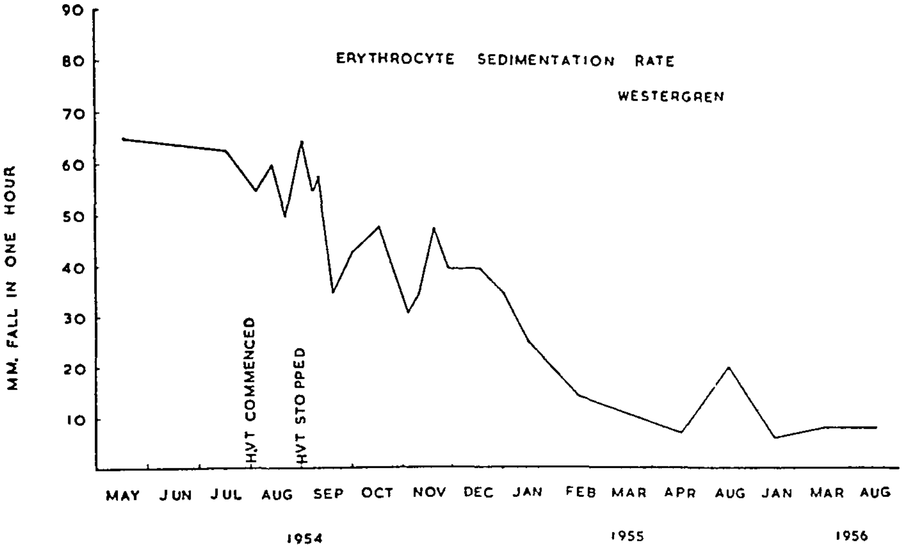


FIG. 7.

Shows how the E.S.R. fell to normal after treatment.

Attempted isolation of a virus by animal injections

Tissue removed at the time of the operation was:

1. Ground with sterile silver sand in a mortar.
2. Mixed with 50 c.c. of sterile normal saline.
3. Passed through a No. 4 Zeitz filter paper.
4. Passed through a Zeitz bacterial filter.

That for intra-cerebral injection was refiltered through a Zeitz filter.

Four mice, two young rats and four rabbits were the animals employed in the experiment.

The mice and rats were injected intra-peritoneally, one rabbit intra-muscularly, and the other three intra-cerebrally, in one of which the material was boiled and cooled before injection.

An ether anæsthetic was administered for the intra-cerebral injections.

At varying intervals after the original injections were made, post-mortem examinations were carried out on 8 of the animals. In none was there evidence of infection either at the site of the injection or elsewhere.

CASE III, a female, aged 41 years.

July 1954. The patient was treated for bilateral maxillary sinusitis by repeated antral lavage and systemic penicillin.

In late July and September 1954 all her remaining teeth were removed and dentures fitted. Very soon she developed pains in the left maxilla, severe enough to disturb her sleep, they radiated into the left temporal, parietal, and post-auricular regions. Associated with this pain there was some difficulty in opening the mouth.

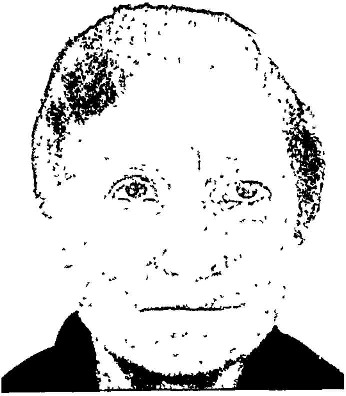


FIG 8

Note the upward displacement of the left eye.

February 1955. Her dentures became difficult to wear because of a swelling, that fluctuated in size, in the left alveolar and cheek regions. This swelling gradually increased and spread to the zygomatic and temporal area.

March. A horizontal diplopia especially when looking to the left was complained of.

April. An X-ray of the sinuses showed the outer antral wall to be destroyed, and the left mandibular condyle to be broader and thicker than usual. Dental X-rays revealed an erosion of the antral floor in the region of the $\underline{|6}$. An X-ray diagnosis of malignant disease of the antrum was made. The patient was then transferred to the ear, nose and throat department.

May 25th. There was a firm and slightly tender swelling involving the left cheek, and a fullness in the left temporal fossa. The left eye appeared to be displaced upwards (Fig. 8). Trismus was severe. The left bucco-alveolar sulcus was obliterated by a firm tender swelling. There was much crusting in the left nostril associated with a peculiar smell.

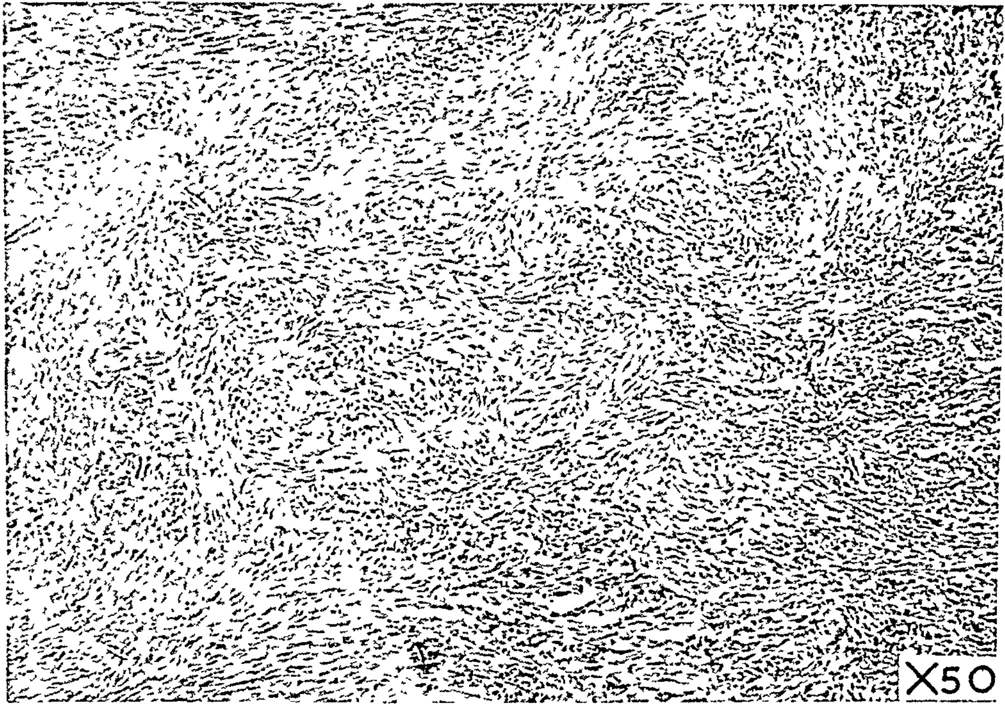


FIG. 9.
A section from the "growth" shows the whorled and densely collagenous material. × 50.

An X-ray of the sinuses showed the right antrum to be opaque, and the outer wall of the left to be completely destroyed. A left antral puncture was performed. The naso-antral bony wall was absent. A large quantity of thin, foul pus was washed out, culture of which produced a heavy growth of *staphylococcus aureus*, coagulase positive, sensitive to all antibiotics.

A wet fixed film was made of a centrifuged deposit but no malignant cells were seen.

May 30th. On admission to hospital a course of 500,000 units of crystalline penicillin twice daily was started for 25 days.

June 1st. Operation under general anæsthesia. An incision was made into the left canine fossa, the blade entered the antrum without difficulty, by cutting through a firm mass which had replaced the outer antral wall. On further inspection inside the antrum, there was a large pale mass attached to the anterior wall, which when cut into, looked and felt like a carcinoma. This material was removed piecemeal, but no attempt was made to perform radical operation, the main aim being to produce adequate drainage for the infection, and to acquire tissue for histological examination. The naso-antral wall, and the outer antral wall were missing. The incision was left open.

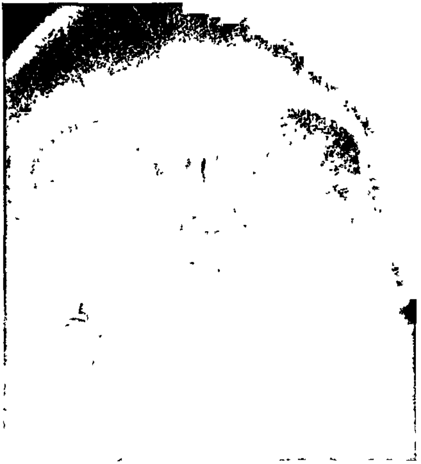


FIG. 10.

Shows destruction of the lateral wall of the left antrum.

Microscopically the tissue removed was fibrous, fasciculated, whorled and densely collagenous in places. The superficial areas were of looser texture, consisting of granulation tissue rich in lymphocytes and plasma cells; no covering epithelium was present. A carcinoma could be confidently excluded (Fig. 9).

June 29th. On the 28th post-operative day, despite the negative histological findings, a course of cobalt 60 was started to the right anterior and left lateral fields, and on August 9th a similar treatment was started to the right lateral field. Over a period of 45 days a tumour dose which varied between 7,026 and 6,100 "r" at its centre, was given.

September 8th. There had been no ill effects from the cobalt therapy, apart from some blurring of vision in the left eye due to a developing keratitis. The swelling in the left temporal fossa which had disappeared at the end of treatment, had returned much larger than before. The trismus was unaltered. The left nasal cavity was dry and atrophic, with some crusting. The oro-antral fistula was patent and healthy, but much smaller.

October 22nd. The swelling in the left temporal fossa had increased in size, and there was a persistent ache on the left side of the head. Further X-rays showed involvement of the left mandibular head, neck and upper part of the ascending ramus; the lateral antral wall was missing (Fig. 10.)

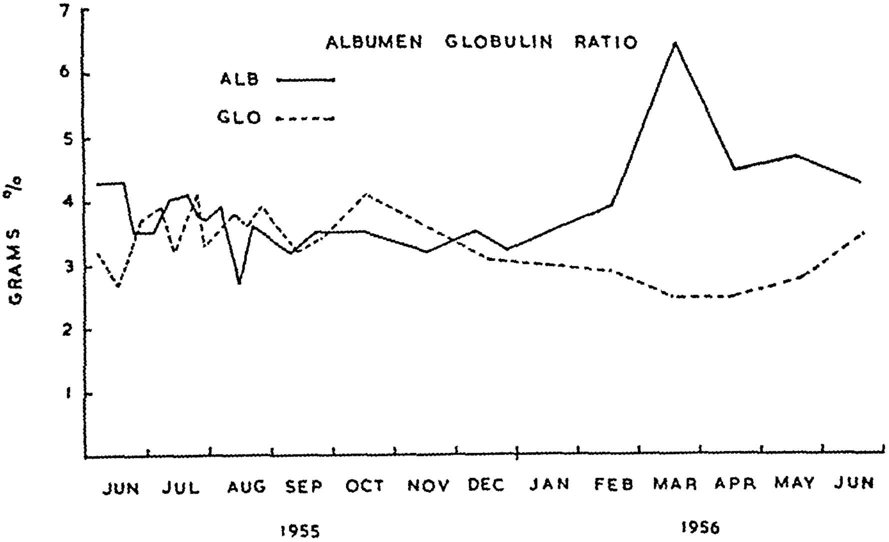


FIG. 11.

Shows the reversal of the albumin-globulin ratio at the height of the disease.

October 30th. A second course of cobalt 60 was started to the same areas, a tumour dose was given which varied between 2,249 and 1,895 "r" at its centre.

March 1956. Her general condition had improved greatly. The swelling in the left temporal fossa was no longer apparent. The vision was still blurred. The nasal cavity was atrophic, but the antrum was healthy.

September 1956. The patient remained quite well. There was no evidence of a recurrence.

Investigations

After the first attendance weekly laboratory investigations were made, later they were carried out monthly. A monthly chest X-ray never showed evidence of disease Urinalyses were always normal Pus from the antrum repeatedly grew *staphylococcus aureus*

Blood The Wassermann and Kahn reactions were negative On no occasion did the blood urea rise above 69 mg per cent The red blood and white blood cell counts and differential white cell counts were always within normal limits There was a mild iron deficiency anæmia

The serum proteins estimated by a salt fractionation method, and the E S R (Westergren) are recorded graphically (Figs 11 and 12) The serum proteins were also estimated electrophoretically

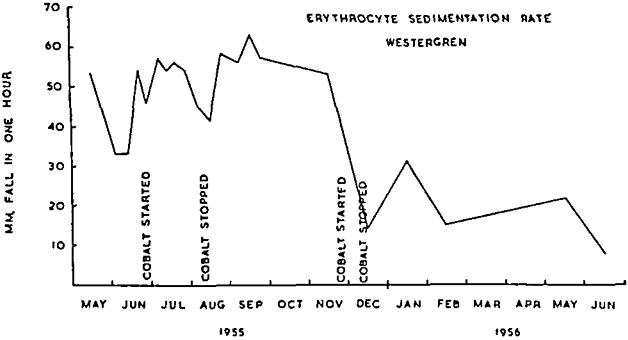


FIG 12
Shows how the E S R fell after treatment

Attempted isolation of a virus by animal injections

The same procedure was carried out as in Case II, but on this occasion four species of animal were used, hamsters and ferrets were chosen because of their marked susceptibility to virus infections

A light ether anæsthetic was administered during the injections of the hamsters, and for intra-cerebral injections in the other species

Approximately 3 months later each animal except the male ferret was killed by chloroform Careful post-mortems were carried out In no animal was there any evidence of infection either at the site of injection or elsewhere

Treatment

Because the ætiology and nature of the disease in early cases was obscure, the treatment was of necessity empirical

A wide variety of local treatments were employed, which ranged from the use of galvanocautery to the application of 25 per cent. chaulmoogra oil. "This", said G. B. Woods, "gave more relief than anything." Douchings with potassium permanganate, weak iodine, sodium hypochlorite, acriflavine, and painting with mercurochrome, the application of cupric sulphate, or packing the wound with urea, all gave no relief.

High caloric diets with heavy vitamin therapy, and on some occasions, transfusions of fresh blood did not help.

Where the surgeon believed the disease to be that of atypical syphilis one or more full courses of the antisyphilitic treatment in vogue at the time was exhibited, always without avail.

Hoover (1941) was the first to use a sulphonamide in the treatment of the disease; this was administered in the form of sulphanilamide. The only observation made was that smears from the lesions grew less streptococci than previously.

Sneddon and Colquhoun, in 1952, reported the cure of a case of granulomatous ulcer of the nose treated with chloramphenicol. This patient died eighteen months later with a recurrence in the pharynx (personal communication).

Penicillin and many other antibiotics have frequently been used without effect. In searching for the correct form of treatment, it is of value to study the methods employed in cases that were alive on reporting.

No fewer than 12 of the 15 cases were treated by some form of irradiation, one by surgery, one cortisone and one corticotropin.

Hoover's was the only case to be cured by excision, it also had a full course of antisyphilitic treatment.

Cortisone was first used by Moore *et al.* in 1950 for the treatment of a fifty-year-old female with a malignant granuloma of the nasal septum, who also had a similar subglottic lesion. Later the patient developed symptoms of rheumatoid arthritis, and skin lesions of disseminated lupus erythematosus. At various times over a period of six months she received a total of 9·5 g. of cortisone. Moore commented: "except for its effect on her glucose tolerance and some possible kidney involvement, she has shown no ill effects from its continued use. When the cortisone has been stopped, the granulomata have increased rather promptly in size, but the last period before relapse was about four weeks as against only a few days previously". At the time of reporting, ten months after treatment commenced, she was being kept on a maintenance dose of A.C.T.H. There were no visible granulomata in the nose, nasopharynx, larynx or trachea. The skin lesions had disappeared.

Hagens *et al.* (1953) were able to present a more dramatic cure. Corticotropin was used to treat a sixty-year-old male with a malignant granuloma of the nose.

"The patient was given 25 mg. of corticotropin intramuscularly four times a day. He was placed on a low salt diet and the intake-output, weight and blood pressure were charted After five days, the same dose of corticotropin was given three times a day for twelve days, and then 17 mg was given three times a day for eight days At about the end of two weeks' treatment a definite improvement in the lesion was noted, and at the end of three weeks the contour of the lesion was almost flat, and the skin appeared to be rapidly filling in The left nasal passage was more open, but crusting was noted over the involved area Use of corticotropin was stopped after 25 days' treatment The facial lesion continued to heal and has remained healed, without further treatment" This patient was alive and well in April 1956 (personal communication)

In 1921, Sir Robert Wood's second case was treated by radium emanations, the patient unfortunately succumbed two and a half years later

Deep X-rays appeared to cure the local condition in M'Arthur's 1925 case The patient died from bronchopneumonia, and was said to have "miner's phthisis" despite repeated negative sputum tests An ulcer on the roof of the mouth treated with 900 "r" by Hoover (1941) seemed to hasten the necrosis, further therapy was therefore abandoned.

Heinermann (1938) was the first to record a cure with X-rays (7,000 "r"). In the summary of a report by Isbell (1945) he said "the use of proteolytic agents, potassium permanganate irrigations and avoidance of trauma, through the use of X-rays, have offered the only worthwhile treatment in the absence of specific therapy"

It was *not until a meeting of the Royal Society of Medicine in 1955 that the results of treatment by X-rays could be evaluated* The most striking observation made in all cases was the small dose required to produce healing

A case described by Howells was still quite well one year and three months after treatment with 1,500 "r".

Gilroy Glass also favoured small doses of X-rays, justifying this by saying "such low dosages could not possibly lead to either sequestration or to irradiation necrosis Moreover, it would be possible to repeat a course of treatment if the condition broke into another area" 50-100 "r" once or twice weekly were advocated Maxwell Ellis was able to report a patient with no sign of recurrence eight years after treatment with 300 "r", this is the longest cure to be found anywhere in the literature.

Sneddon's second case (unpublished) was a woman of twenty-nine who had a recurrent granulomatous ulceration of the central face for about six years All forms of chemotherapy had failed to check the process She was ultimately treated by 2,000 "r" of superficial X-rays which caused healing

In a later paper, Paterson (1956) presented two cases in which healing had taken place after treatment with X-rays, one in combination with antibiotics and cortisone, the other X-rays alone He believed that the administration of antibiotics at the time of X-ray treatment gave the best results.

Because the clinical appearances so closely resembled that seen in carcinoma of the antrum, Cases II and III were treated with cancercidal doses. The main difference being the use of conventional high voltage therapy in Case II, and cobalt 60 in Case III.

A tumour dose of 4,555 "r" was given over a period of 30 days in Case II.

The first course in Case III varied between 7,026 and 6,100 "r" at the centre of the tumour given over 45 days. A suspected recurrence necessitated a second course three months later. Over 25 days the centre of the tumour was treated by a dose varying between 2,249 and 1,895 "r".

For purposes of comparison between the two methods employed 1 "r" in Case II equals 1·3 "r" in Case III.

Prognosis

That the word lethal is so often used in the title when this disease is reported, indicates the commonest outcome. The truth of this statement is well illustrated by the following figures.

Out of a total of 86 collected cases, 60 have died with the disease. Of the remaining 26, 10 are alive but still have the disease; all trace has been lost of one.

Where a five-year cure rate is sought, which is the minimum time allowed in cases of carcinoma if figures are to have any significance, only one, that of Maxwell Ellis, has survived eight years. Of the remainder, 4 are alive and well 2-3½ years, and 10 cases from a few months to eighteen months after completion of treatment.

CAUSES OF DEATH IN 60 CASES

	No. of cases
Cachexia	29
Pneumonia	9
Uræmia	8
Septicæmia	4
Hæmorrhage	3
Cardiac failure	2
Intestinal obstruction	1
Peripheral neuritis	1
Agranulocytosis	1
Sarcoma cutis	1
Miner's phthisis	1

Malignant Granulomas of Nose and Paranasal Sinuses

PART II

Ætiology

There are nearly as many theories about the cause of the disease as there are recorded instances of it

It is possibly by surveying the suggestions put forward in the literature to divide them into three main groups

1 Neoplastic
2 Infective
3 Hypersensitivity

Neoplastic

Few authors subscribe to this theory despite the destructive and lethal nature of the disease

Bergqvist and Koch believe that many cases reported in the literature, as well as their own cases were neoplastic, the histological picture being concealed by infection They say "there are further strong reasons for assuming a malignant new formation both on the grounds of progression of the process, which was that of a malignant tumour, and because in a number of cases it was possible to demonstrate microscopical structures, strongly resembling those of sarcoma There is therefore every reason in these cases of granuloma gangrenescens and malignant granuloma to assume that a malignant new formation is present".

Infective

Because of the close similarity, both clinically and histologically between malignant granuloma and other chronic infective granulomas, the early authors confined their attentions to theories of infection Nearly every case had repeated Wassermann and Kahn reactions performed In many, animal inoculations were carried out in an attempt to isolate the tuberculous bacillus Despite the negative findings, antisyphilitic treatments were persisted in, always without avail Once it became obvious that the disease was not tuberculosis or syphilis a wider search was made for the rarer forms of infection In McBride's case, fluid was drawn from the patients' hydrocele, some of which was injected into a guinea-pig. Secretions from the ulcer were injected into a second guinea-pig Neither animal presented any evidence of orchitis, the characteristic changes produced in these animals by glanders

Stewart says that from the clinical and microscopic appearances the disease is not one of a tumour but essentially a pyogenic one, a chronic inflammatory process

He observed that in six of his ten cases, a streptococcus had been found in association with a staphylococcus, and in the seventh a staphylococcus alone He quoted Brewer and Meleny's theory that the ætiology of this condition may be a symbiosis of these organisms. This theory has been supported on a number of occasions by other authors. Hoover added that the symbiosis may produce a necrotizing substance capable of such destruction. He also suggested that virus studies might be done.

Lewy had a suspicion that a fungus infection was the cause in his case, but repeated examinations failed to prove it.

Hargrove *et al.* state that the process can be best explained by two pathological changes occurring simultaneously: firstly, a locally-spreading lesion, the cellularity of which indicates the property of new tissue formation, whether this abnormal tissue be granulomatous or neoplastic; and secondly, an obliterative arteritis to which can be attributed the fibrinoid degenerations of connective tissue, the accompanying small hæmorrhages, and the complete destruction of more distant tissue.

Sneddon and Colquhoun suggested that the granulomatous overgrowth is a non-specific reaction to a staphylococcal infection and pointed to the common pyogenic granuloma as a good example of exuberant granulation tissue stimulated by such infection. In their case a staphylococcus was repeatedly isolated.

Agglutination reactions for typhoid, paratyphoid, tularæmia, etc., sporotrichosis skin tests; Frei's test for lymphogranuloma inguinale and Strauss test for glanders have been done on many occasions, and were always negative.

Hypersensitivity

Friedmann (1955) believes there are probably two types of malignant granulomas.

1. The classical type with external ulceration of the face; no giant cells in the often necrotic tissue, death being due to cachexia or hæmorrhage.
2. Without external ulceration, destruction usually spreading underneath the surface; often with giant cellular histology. Development of ulcers of the palate, tongue, larynx, and trachea, with widespread necrotizing angeitis, and granulomatosis involving kidneys, spleen, lungs, adrenals and other sites. Death due to uræmia caused by renal failure.

Lesions identical in every way to periarteritis nodosa have been found at post-mortem in cases suffering from serum sickness and treated with sulphonamides, Rich (1942a and 1942b). Experimental evidence has also been given by Rich and Gregory (1943) to show that periarteritis nodosa is a manifestation of hypersensitivity. They say that widely different types of sensitizing antigens are capable of causing periarteritis nodosa in man, and suggest the advisability of attempting to discover and eliminate the responsible antigen in each case diagnosed clinically.

If it is assumed that malignant granulomas are a localized form of periarteritis nodosa, a search has to be made for some factor either chemical or bacterial that is common to all cases

It is possible to exclude the antibiotics as chemical agents in many cases because the disease was described before the introduction of the sulphonamides Trauma, or previous operations to the respiratory tract was frequently mentioned in the histories

If it is not possible to isolate some external agent, then would it not be feasible to incriminate one of the normal inhabitants of the nose?

Non-pathogenic staphylococci and streptococci are found in the nose and nasopharynx of many healthy individuals When studying the bacteriological reports of cultures taken from cases of malignant granuloma, it was found that at least two-thirds of the cases grew a staphylococcus, and even more a streptococcus as well Many gave a history of prolonged upper respiratory infection That such a high percentage of staphylococci is unusual is apparent when only 30 per cent. of one hundred cases of chronic antral infection revealed a staphylococcus as the causative organism.

With these facts in mind it would appear that a staphylococcus or streptococcus or both, were by far the most common agent discovered in these cases It could be argued that in any destructive lesion of the nose or antrum, one of these organisms is most likely to be found, but one would also expect to find other secondary invaders, which, in fact, were rarely found

If a staphylococcus or streptococcus is to blame for this peculiar granuloma, how does it stimulate the body to react in such a way?

In responding to the environmental stress produced by the invasions of micro-organisms, Forbus stated the body makes use of two stereotyped defence mechanisms, suppurative and granulomatous inflammation

Arthus and Breton demonstrated that if an area of skin is sensitized and then an antigen is injected into the area, a pathologic picture similar to that produced in granuloma will result

Williams (1949) says that in idiopathic granuloma, the lesion presents the clinical and pathological appearances of a granuloma, in other words, the normal defence mechanism of granulomatous inflammation, but he is unable to discover a normal stimulus to initiate such a reaction The idiopathic granuloma may therefore be considered an abnormal reaction, and therefore an allergy

The histological picture described by Arkin in cases of periarteritis nodosa, is strikingly like that described by Cannon and Pacheco where they injected a living *staphylococcus aureus* into a previously sensitized guinea-pig and produced a granulomatous lesion

If it is presumed that the destructive ulceration of a malignant granuloma is either a localized form of periarteritis nodosa or the precursor of a generalized form of periarteritis nodosa, then it seems not unreasonable that a staphylococcus, streptococcus, or both are responsible in some way for the development of these granulomatous lesions. It would be more logical to blame an organism that has been shown to be present on many occasions than to say, because no agent could be found, then the reaction is abnormal, and must be allergic.

Pathology

Histology

The words non-specific granulations have been used practically without exception by authors reporting the microscopical findings from their cases of malignant granuloma. In some there was an excess of plasma cells, in others eosinophils, and in a few multi-nucleate giant cells were found, but "granulations" was the only common denominator in all reports.

These facts alone point to the histological picture to be expected.

Stewart (1933) said it was a chronic inflammatory process, with proliferation of the endothelial cells, lymphocytes, and plasma cells and formation of granulation tissue, at first cellular but later becoming fibrous. He stated that material from early cases showed principally round-cell infiltration, with necrosis of the epithelium. As the disease progressed fibrous tissue was laid down, fairly loosely just beneath the surface, and more densely in deeper parts, with small round-cell infiltrates still present amongst the fibrous tissue. The blood vessels were proliferating in the early stages, but later had enormous thickening of their walls, and end-arteritis with hyaline changes.

Little has been added to this description until Friedmann's paper (1955).

He pointed out that the histological picture showed considerable variation and one or the other morphological elements predominated, producing these pictures:

(*a*) non-specific pleomorphocellular inflammatory granulation tissue;

(*b*) non-specific inflammatory granulation tissue with histiocytes predominating;

(*c*) non-specific inflammatory granulation tissue with necrotic changes predominating;

(*d*) non-specific inflammatory granulation tissue with giant cells predominating.

He adds that "several cases, however, presented a varied mixture of all the above-mentioned components, and occasional giant cells were frequently seen".

In my three cases there were no histological grounds for diagnosing more than active antritis in the first A second biopsy in this case consisted mainly of granulation tissue, which in places was densely collagenous There were similar microscopical findings in Case II The macroscopic appearance of the tumour in Case III differed from that in Case II, being whiter and firmer

Histological evidence of a sarcoma, or carcinoma could not be found in any of these three cases, or of the many recorded in the literature The picture is always that of granulations in varying stages of maturity, which is noticeable, not only in different cases, but also in the same mass of tissue In the early stages round cells, polymorphs, and particularly plasma cells, are in evidence, as the disease progresses more and more fibrous tissue appears, which is whorled and fasciculated, especially in the deeper areas

In the three cases reported, no giant cells were found after a very thorough search

It is difficult to give a more detailed histological description than to say that the tissue is very similar to normal granulation tissue which has been replaced by fibrous tissue interspersed with inflammatory cells, the number diminishing with the increase in maturity

Blood Picture

There is usually little or no change in the blood counts, on rare occasions the total white blood count may rise to 15,000 per c mm The differential count is nearly always within normal limits The close association of this disease to the classical periarteritis nodosa might lead one to expect a rise in the eosinophils In my three cases together with those in the literature without signs of periarteritis nodosa, there was never any significant rise in the eosinophils

Geist and Mullen (1953) report their first case as having 18 per cent eosinophils, an autopsy revealed an acute necrotizing arteritis involving many organs of the body

Howells and Friedmann (1950), Stratton *et al* (1953), and McCallum (1954) were unable to find a rise in the eosinophils in any of their cases of granuloma of the nose associated with periarteritis nodosa

The haemoglobin tends to fall as the disease progresses, probably secondary to toxic absorption from the necrotic granulation tissue

Blood Urea

It is interesting to note that uræmia has been cited as the cause of death in no fewer than 8 cases, all of which had periarteritis nodosa This fact alone indicates the value of estimating the blood urea as an index of kidney function in these cases

Urinalysis

A significant quantity of red blood cells, casts, and albumin in the urine may indicate the involvement of the kidneys in a diffuse type of periarteritis nodosa.

Sedimentation Rate

The sedimentation rate is measure of the presence and intensity of morbid processes within the body. One of the most important uses of the E.S.R. is in calling attention to the presence of a more or less occult disease. Other things being equal, an accelerated rate suggests organic disease rather than a functional disorder, an inflammatory condition than a tumour, and malignancy than a benign tumour.

Whitby and Britton say: "The main factor for producing an increased sedimentation rate, as well as the most important from the practical aspect, is the extensive destruction of body cells, which occurs in infections and toxic conditions."

In Cases II and III the E.S.R. was found to be very high during the active stage of the disease, only falling after treatment. It became obvious that E.S.R.s in cases of malignant granuloma were of immense value in judging progress of treatment. This did not, however, answer the question why the E.S.R. was so abnormally raised in what appeared at first sight to be a very localized condition.

In practically all recorded cases of malignant granuloma there is a very heavy superadded infection. With the knowledge that infection and tissue destruction are responsible for an acceleration in the E.S.R., 25 cases of purulent maxillary sinusitis had E.S.R.s estimated, in only 2 did the reading reach 20 mm. fall in the first hour (Westergren), of the remainder none were higher than 8 mm. The results of this investigation are further borne out by the findings of Ballantyne and Rowe (1949), who estimated the E.S.R. in 206 consecutive and unselected cases of chronic maxillary sinusitis and noted that the readings were always within normal limits.

From these findings it was apparent that infection alone was not responsible for the high E.S.R. It was therefore felt that proven cases of malignant disease of the antra with superadded infection would be more comparable to cases of malignant granuloma.

Ten cases of malignant disease were selected because of the severe destructive nature of the lesions, and their associated infections. In 8 cases of poorly differentiated carcinoma involving the maxillary sinus, and ethmoids, two of which had discharging fistulae at the inner canthus, the E.S.R. was estimated; in only 2 did it rise to 33 mm. fall in the first hour, the others ranged between 15 mm. and 25 mm. In two patients with lymphosarcoma, one had an E.S.R. of 22 mm., and the other only 3 mm.

Having exhausted the possibility that these high readings were due to infection, or tissue destruction, or both, a further search had to be made for a cause

Increase in the E S R. is very marked in severe acute anæmias, the rate increasing rapidly as the red cell count decreases There being no anæmia in Cases II and III, this cause was discounted

A search of the literature shows that the very few E S R s estimated in cases of malignant granuloma have practically all been raised There has been no previous attempt at consecutive recordings to help judge progress of the disease

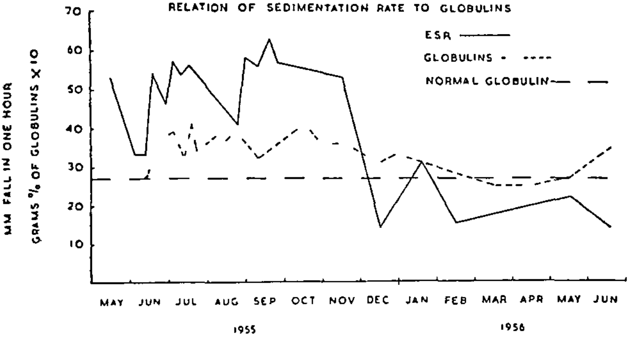


FIG 13

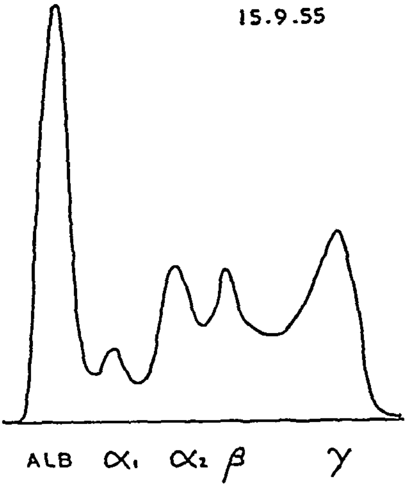
Shows how the E S R and the serum globulin return to normal as the disease is cured

An attempt to elucidate the cause of the high E S R in Case II was made It has been known for many years that a high E S R is usually due to an increase in the concentration of the fibrinogen, or globulin, or both, in the blood The origin and significance of such quantitative changes may of course vary In infectious diseases, antibodies of globulin nature probably increase the globulin fraction, and the infectious agent may stimulate the production of fibrinogen

With this in mind the serum proteins were estimated The fibrinogen content was within normal limits, but a very significant finding was the reversal of the albumin-globulin ratio, when estimated by a routine salt fractionation method As the clinical condition improved, the E S R fell, and the albumin-globulin ratio changed to normal, indicating that at least in cases of malignant granuloma the increase in globulins might be responsible for the high E S R readings

There were similar findings in Case III (Fig. 13); but of greater interest were the findings by electrophoresis. At the height of the disease there was found to be a marked reversal of the albumin-globulin ratio, with an unusually high gamma globulin level (Fig. 14).

Ropes *et al.* (1954) state that consecutive observations of the electrophoretic distributions of plasma proteins during the course of exacerbations and remissions in rheumatoid arthritis indicate that the protein changes reflect alterations in clinical activity of the disease, and correlate in general the variations in the E.S.R.



	15.9.55	*Normal*
Albumin	26·27 %	58·62 %
Alpha 1. Globulin	6·24 %	3·5 %
Alpha 2. Globulin	13·17 %	9·11 %
Beta Globulin	14·58 %	11·13 %
Gamma Globulin	39·75 %	12·16 %

FIG. 14.

Is an electrophoretic graph showing the increase in gamma globulins at the height of the disease in Case III.

Malmros *et al.* (1946) say "it is evident that a high sedimentation rate may be due to a rise in either alpha, beta or gamma globulins, or to some continued increase thereof".

Once the possible cause of the high E.S.R. in malignant granuloma had been ascertained, it was felt that further investigations into the globulin subfraction changes might be of value in revealing more of the nature of this disease.

Plasma Proteins

As yet there is no universally accepted technique, or findings of the electrophoretic methods of protein estimation, but most authorities, no matter what method they employ say that the globulins can be divided into a variety of subfractions Where figures for health and disease have been published most agree that one or other subfraction is raised or lowered for any one specific disease

Flynn states "the usual significance of the changes in the electrophoretic globulin fractions may be briefly summarized as follows

(*a*) the alpha globulins are increased in high fever, tissue destruction, nephrotic syndrome (alpha 2) only

(*b*) the beta globulin is increased in conditions associated with increase of the phospholipids and cholesterol, myeloma (some cases)

(*c*) the gamma globulin is increased in chronic infections, liver disease, collagen disease, myeloma (some cases)

The most notable and consistent feature in collagen disease, even in the early stages, is a considerable increase in the gamma globulin". Periarteritis nodosa is classified as a collagen disease

Whilst the literature on electrophoresis in many diseases is abundant, data concerning ear, nose and throat disease is scarcely to be found

Surján has collected 16 cases of carcinoma of the larynx, and 9 cases of maxillary sinusitis in which he has estimated the serum proteins by electrophoresis In none did he find any significant change

Rarely were the serum proteins estimated in cases of malignant granuloma reported in the literature, in one of Levan's cases there was a raised globulin, and in the other a marked albumin-globulin reversal, as was also shown in McCallum s case

The electrophoretic patterns in Case III revealed a staggering increase in the gamma globulin, and also a very marked increase in the alpha 1 globulin which tends to rise where tissue destruction is present That these findings are so specific for a collagen disease does lead one to suspect that malignant granuloma should be classed in this category and not in an infectious, carcinomatous, or sarcomatous group

Bacteriology

Of all the investigations carried out on cases of malignant granuloma, the most thorough has been bacteriological

Many people have performed routine cultures of smears taken from the diseased area, additional laboratory investigations have been used to try to find some organism responsible for the disease Nearly every known agglutination test has been carried out at one time or another, and always with negative results The Wassermann and Kahn reactions on the serum were done in nearly every case, in none was a positive result returned Animal inoculations, cultures, and Mantoux tests have failed to show any relationship to tuberculosis Careful searches for spores and fungi have been without avail

It has been tentatively suggested that a virus might be a responsible agent, but no real effort has been made to isolate it. In Cases II and III as recorded earlier, such tests were carried out with negative results. It is not claimed that this small investigation in itself excludes a virus infection, but at least shows that a wide variety of laboratory animals appear to be unaffected when injected by a bacterium free filtrate of malignant granuloma tissue.

If further cases become available a useful line of investigation would be the injection of bacteria from the diseased area, and also bacteria-free filtrates from the "granulations", into the nasal mucosa of laboratory animals, and possibly into some that have previously been sensitized by frequent small injections of dead staphylococci.

It is only when all the published figures on bacteriology are studied that some light is thrown on the possible cause of the disease.

Out of 86 cases of malignant granuloma, 37 had a bacteriological investigation carried out on smears taken from the lesions. In no fewer than 35 of these a staphylococcus was isolated, 12 of which grew a streptococcus as well, a streptococcus was found alone in 3, and a pneumococcus in one of the remaining 4 cases. This analysis reveals an unusually high percentage (89 per cent.) of those investigated to have a staphylococcus present. When a search is being made for some common factor in each case, this finding is very significant.

Discussion

After conversations and correspondence with many ear, nose, and throat surgeons, and reading the relevant literature one is lead to believe that the condition of malignant granuloma is not as rare as reported figures suggest. Lesions in the nose or antrum are occasionally treated as carcinomata on clinical evidence alone, despite one or more histological studies to the contrary. May not some of these cases be malignant granulomas?

It might be said that there are too few cases recorded on which one can make dogmatic statements, and that figures are not large enough to be statistically significant; however, until some better understanding of the disease is forthcoming, and an efficient remedy devised, the only consolation about this condition is its apparent rarity. It does not seem rare to the patient who has it, or to his bewildered family.

By searching carefully into the medical histories, investigations, treatments, and outcome of many reported cases of malignant granulomas, and by a thorough investigation of two out of three personal cases, new features have either been brought to light, or those already noted, clarified. It is proposed to try to show with the aid of some researches recorded earlier, that the condition of malignant granuloma should be classified as a collagen disease, rather than an unusual form of carcinoma, sarcoma, or even a simple infection as some authorities believe

The histories of Cases I, II and III are more or less the same, resembling closely those recorded in the literature, the first died despite many forms of treatment It is only when studying this case in retrospect that two findings of interest are noted It is possible that the abdominal pain and mass in the right iliac fossa were manifestations of a diffuse periarteritis nodosa, unfortunately a post mortem could not be performed to verify this suspicion The other interesting observation was the unusually high E S R for such a localized disease

The value of estimating the sedimentation rate is very well illustrated in Cases II and III In both it was used to judge the progress of the disease, and the effect of treatment It can clearly be seen on the graphs, particularly in Case II, how the sedimentation rate fell to normal as the local condition was cured In Case III a sudden rise after a gradual fall indicated a possible recurrence of the disease This finding was borne out a week later when a recurrence became clinically evident

A further valuable use of the E S R was also shown in this case The patient was diagnosed clinically and radiologically as a carcinoma of the maxillary antrum The E S R was done and found to be higher than 50 mm fall in the first hour A malignant granuloma was immediately suspected knowing that carcinomas sarcomas or simple infections rarely gave rise to such a high fall

The macroscopic appearance of the tumour seen at operation closely resembled a carcinoma It was not until the histology had failed to show a carcinoma, but a malignant granuloma that the great value of the E S R , in these cases was fully realized

It is therefore suggested that in disease of the nose or antrum where a malignant condition is suspected and the histology does not verify this suspicion, an E S R would be a very valuable test to perform If the fall in the first hour was greater than 50 mm when corrected for anæmia, and there were no other signs of disease present, it would strongly suggest a malignant granuloma rather than a carcinoma or sarcoma Once a diagnosis of malignant granuloma has been made, it is further suggested that a weekly E S R should be performed to aid clinical observations in judging treatment and progress

It has been known for many years that alterations in the blood globulins and fibrinogen were directly related to changes in the sedimentation rate Investigations into the causes of the high sedimentation rate in Case III revealed some startling changes in the blood proteins The normal salt fractionation methods showed a reversal of the albumin-globulin ratio, with a normal fibrinogen level This pointed to the globulins as being the responsible factor in this case It was not until these globulins were split up by electrophoresis that a greater significance was attached to this finding. It has long been suspected that malignant granulomas were in some way related to periarteritis nodosa. Fig. 15 shows the age incidence in 300 cases of periarteritis nodosa, and 86 cases of malignant granuloma; the neoplasms were of the antrum and nose that died in Great Britain in one year (Registrar-General figures). The graph illustrates how closely the two diseases resemble each other in age incidence.

Apart from clinical observations, no one has been able to find a common factor in the two diseases. The isolation of a very high gamma globulin level in this case of malignant granuloma was identical to one of the most consistent blood changes in collagen disease in particular periarteritis nodosa.

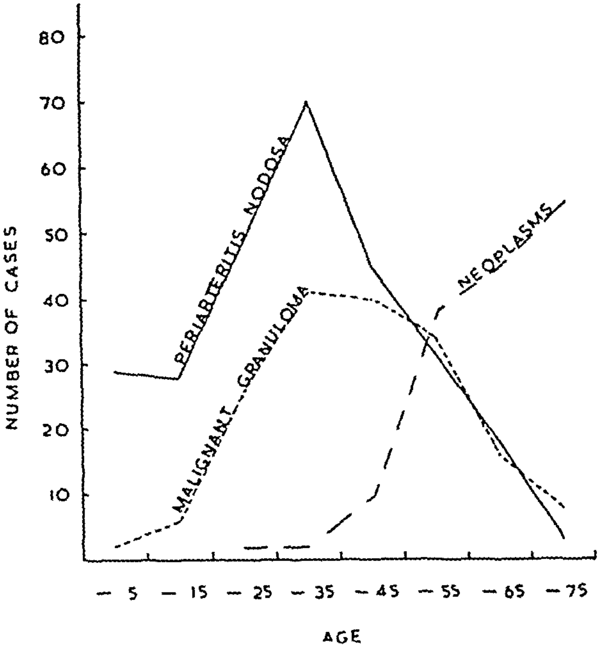


FIG. 15.

Shows how closely the age incidence of periarteritis nodosa and malignant granulomas resemble each other.

Cecil and Loeb say: "The term 'collagen disease' is not a diagnostic symbol which identifies a specific morbid entity, well defined by characteristic clinical features, or post mortem tissue alterations. It rather refers to a concept founded upon pathologic-anatomic observations which suggest that clinically heterogenous maladies have a common denominator in generalized alterations of the connective tissue, specifically of its intercellular components."

Friedmann, when discussing the pathology of malignant granulomas, felt they should be divided into two main groups.

(*a*) "The classical type with external ulceration of the face, no giant cells in the often necrotic tissue, death being due to cachexia or hæmorrhage No polyarteritis is found at post mortem "

(*b*) "Without external ulceration, destruction usually spreading underneath the surface Often a giant cellular histology Development of ulcers of the palate, tongue, larynx, trachea, with a widespread necrotizing angeitis, and granulomatosis involving kidneys, spleen, lungs, adrenals, and other sites Death due to uræmia caused by renal failure "

This division is based on clinical and histological findings alone If the E S R s and blood proteins were estimated in these cases, it would probably be found that type (*a*) produced a systemic response identical with that of type (*b*), thus indicating that the first variety is a localized manifestation of the second It is possible that if cases of the localized disease lived long enough it would develop signs of diffuse periarteritis nodosa

Although Case III is the only one recorded in which electrophoresis has been used to isolate the globulin subfractions the findings, together with the high E S R , the known clinical relationship, and the age incidence do suggest very strongly that malignant granulomas are one of the clinical manifestations of periarteritis nodosa

As with the E S R , the blood globulins especially when estimated by electrophoresis, are of great value in both diagnosis, and judging the course of the disease Blood proteins show only slight variations in cases of carcinoma, or sarcoma If a suspected case of carcinoma revealed a very high E S R and a reversal of the albumin globulin ratio, with an unusually high gamma globulin level a diagnosis of a malignant granuloma should be the first choice

Routine urinalysis, blood urea estimations, and frequent chest X-rays should be performed in cases of malignant granuloma These may reveal one of the first signs of a diffuse periarteritis nodosa

In searching for a cause of malignant granuloma a very limited virus investigation was carried out Although it was realized that the use of human tissue cultures would have given more information, it is felt that the injection of a cell and bacteria free filtrate of the tissue extract into animals, particularly through the intracerebral route, were of value, inasmuch as these animals were unaffected by the virus, if indeed one was present at all

One finding common to these three cases and to many others in the literature was the isolation of a staphylococcus in cultures grown from the lesions Histologically malignant granulomas in no way resemble one of the usual neoplasms in the nose or antrum or even a normal inflammatory reaction to infection However, the ' granulations" must have some causative agent It would appear possible that a bacterium, such as a staphylococcus, could evoke an unusual response in the form of a hypersensitivity reaction. In tissues previously sensitized to a staphylococcus, it is possible to produce a granuloma by injection of a staphylococcus into the area; such a response would be a hypersensitivity reaction. One can visualize this process taking place on a much larger scale in the presence of unlimited staphylococcus antigen inside an infected antrum. Masses of "granulations" would be produced as a wave of hypersensitivity spread through all the tissues. This could serve as an explanation of the destructive nature of malignant granulomas. It is apparent from clinical observations that the granulations are never extensive enough to cause destruction of bone by pressure alone. The way the disease spreads, destroying all before it, is very obvious once the skin is involved.

If this hypothesis is correct, a staphylococcus, streptococcus, or an organism as yet unidentified, may be held responsible for the initial hypersensitivity, and later the production of masses of "granulations". It is interesting to note that many of the cases gave a history of a previous upper respiratory infection. It may be that Stewart's prodromal stage in which there is a stuffiness of the nose accompanied by a watery nasal discharge, is the early stage of a hypersensitivity reaction, and only when the "granulations" have formed do the patients present themselves for treatment.

No specific treatment is known for malignant granuloma at present. Where successes have been recorded it is nearly always following the use of irradiation.

If the disease is due to an abnormal reaction to bacteria it would explain why such small doses of X-rays as advocated by Howells, Gilroy Glass and Maxwell Ellis have been so effective in producing healing in some cases. Those that have been apparently cured with cancercidal doses might have done equally well with a smaller dose.

If a localized form of malignant granuloma is found, the best treatment would appear to be adequate drainage procured at the time of biopsy in the case of antral lesions, together with a prolonged course of the appropriate antibiotics.

Irradiation should be given producing approximately 2,000 "r" at the centre of the tumour. The progress of treatment being judged clinically, and by frequent E.S.R. and blood protein estimations. Routine urinalysis, blood urea, and chest X-rays should also be employed to indicate any sign of spread of the disease. In this way, the apparently rare malignant granulomas may be cured.

Summary

The literature on malignant granulomas is fully surveyed, three personal cases are reported. In using these cases and facts collected from

the literature, new evidence has been produced to justify the inclusion of malignant granulomas as a "collagen disease"

Virus studies were carried out on tissue removed at operation, these were of value in showing that the usual laboratory animals were unaffected by injections of a bacterial free filtrate into various sites

Routine bacteriological investigations repeatedly revealed a staphylococcus, this being the commonest organism isolated by other authors The finding of a staphylococcus more frequently in cases of malignant granulomas, than in normal noses, or in infected antra suggests that they may have some ætiological significance A theory has therefore been postulated in which a hypersensitivity reaction to a staphylococcus or streptococcus produces masses of granulation tissue giving rise to the destructive condition of malignant granuloma

The value in using the E S.R as a diagnostic aid, and in judging progress of treatment has been well illustrated in Cases II and III, they being the only ones recorded where this test has been employed routinely

An entirely new feature of this condition has been observed following the use of electrophoresis in estimating the blood proteins It was found that there was a marked reversal of the albumin globulin ratio, with an excessively large amount of gamma globulin this being a common finding in cases of periarteritis nodosa

A method of treatment using a combination of surgery and irradiation is advocated.

Acknowledgements

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RAPID HEALING AFTER MASTOID SURGERY BY THE USE OF THE POST-AURICULAR FLAP

By PHILIP H. BEALES (Doncaster) and WILFRED HYNES (Sheffield)

In 1938 Lempert popularized the fenestration operation when he described a one stage procedure, and this operation is now firmly established in the surgical treatment of otosclerosis. More recently new techniques have been described by Zöllner and Wüllstein which enable the otologist not only to remove the disease completely in chronic otitis media, but also, by means of a plastic procedure, to reconstruct a new tympanic cavity in such a way that the hearing is improved and is not destroyed as is usual with the classical radical mastoidectomy. The radical mastoid operation, although it may still have a place in otology, is rarely indicated today.

Though the techniques of these operations are now becoming standardized and are satisfactory as far as the middle ear is concerned, they are all surgically unsound in that they produce raw areas without epithelial cover; these are the result of the surgery of access through the mastoid process and consist largely of exposed mastoid bone. If no effort is made to cover these raw surfaces, they either remain unhealed and result in permanent discharge, or they heal spontaneously in time after careful daily toilet. Apart from delay, spontaneous healing is apt to produce an unstable epithelial cover which tends to break down with the passage of time.

Efforts have been made to expedite healing by covering the exposed mastoid bone with a split skin graft, but this has also proved disappointing. The satisfactory initial "take" of a skin graft on exposed mastoid bone can often be obtained but unfortunately complete or partial breakdown of the graft usually occurs after an interval, which may be as long as twelve months in the case of a grafted fenestration cavity. The problems of healing of mastoid cavities was discussed at a recent meeting of the North of England Otolaryngological Society (1957) and it was concluded that no completely satisfactory method was yet available, although Thorburn was of the opinion that small pinch grafts gave better results than split skin grafts in the mastoid cavity. One of the authors (P.H.B.) demonstrated the behaviour of split skin grafts in the mastoid cavity, and in previous publications has shown skin sections taken from the mastoid cavity after several months had elapsed (Refs.). It was then demonstrated that skin grafts placed in the mastoid cavity underwent atrophy and disintegration after an initial "take" and were replaced by granulation tissue which became infected, leading to discharge which was often impossible to eradicate.

It has been said that there is "nothing more gratifying to the surgeon or to the patient than a successful fenestration operation; but there is sometimes, unfortunately, a heavy price to pay, for the operation cavity may discharge persistently, even for years" (Ref. *Lancet*, 1957). Simson Hall, in his Presidential address to the Section of Otology, Royal Society of Medicine, in November 1957, stated that 13 per cent. of his patients after fenestration had a persistently discharging cavity. Terence Cawthorne in a personal communication (1958) states that at least 15 per cent. of his fenestration cavities are discharging at any one time. Simson Hall also says, "there is no doubt in my mind that the fenestration operation produces a life-long disability in some patients and potentially in all; further there are certain occupations or amusements which are barred for good to the patient, bathing is always a potential source of danger, and surfing and diving are absolutely forbidden".

It is the disability produced by the large vulnerable mastoid cavity that has led workers to experiment with the closed fenestration operation, a procedure which does not give such good results. It is also possible that the present interest in the Rosen operation would be less enthusiastic if a technique for fenestration could be devised which abolished the possibility of discharge from the mastoid cavity.

Tables of Healing Time after Mastoid Operations

A. *Fenestration Operation*

I. B. Thorburn (personal communication)

TABLE I.

Thirty six Cases with Full Thickness Grafting of Mastoid Cavity.
April 1955–November 1957

Time	6 weeks	12 weeks	6 months	1 year
Healed	8	13	17	17
Moist	9	9	6	8
Discharging	19	13	8	2
No Information	—	1	5	9

TABLE II

Thirty-six Cases without Skin Grafting of Mastoid Cavity
May 1953–February 1955

Time	*6 weeks*	*12 weeks*	*6 months*	*1 year*
Healed	18	29	32	33
Moist	—	4	3	3
Discharging	18	3	1	—
No Information	—	—	—	—

These figures show that although healing appears to occur more rapidly if the cavity is grafted, at the end of 12 months less than 56 per cent. of the cases are in fact healed if the cavity has been grafted with a free graft, but over 90 per cent. of cases are healed if the cavity has not been grafted.

Thorburn comments that "these figures do not give a true picture of the final state of healing since quite a fair proportion have been inclined to relapse to an unstable condition".

R. B. Lumsden (*J. Laryng.*, February 1958)

TABLE III.

HEALING TIME OF 148 FENESTRATION OPERATIONS.

	Number of operations	Average healing time in weeks	Recurring infection
No Thiersch grafting of cavity	120	20·5	25·5%
Primary Thiersch grafting of cavity	28	8·8	32%

B. *Healing Time after Tympanoplasty Operations* (*excluding myringoplasty*)

P. H. Beales

TABLE IV.

162 CASES WITH NO SKIN GRAFTING OF MASTOID CAVITY.

Number of operations	Healing time	Incomplete healing of mastoid cavity
162	10 weeks to 6 months	20%

I. B. Thorburn

TABLE V.

SIXTY-THREE CASES WITH FULL THICKNESS GRAFTING OF MASTOID CAVITY.

Healing time	8 weeks	16 weeks	6 months
No. of operations: 63 Healed	28	47	52
Failed to heal: 11 cases or 19%			

These figures show that about 20 per cent. of cases fail to heal and that grafting of the cavity has little effect on the ultimate healing. They do not take into account the later breakdown of the apparently well healed cavity and this occurs, in my experience, more frequently if the cavity has been grafted

C *Healing after Radical and Modified Radical Mastoid Operations*

F McGuckin, in a personal communication (1958), states that in 1951 or 1952 he was able to discuss 650 cases which had undergone radical or modified radical mastoid operations in which accurate records of healing time had been kept Over a period of three years he managed to get the healing rate to about 80 per cent within six months A strict follow-up showed that of these healed cases one in three broke down again within a year

Comment on Figures of Healing Times after Mastoid Operation

These figures represent a large number of cases followed up over a period of years by several workers

They clearly show that about 20 per cent of all mastoid operations fail to heal completely They do not show the number of cases which although apparently well healed break down again and there can be no doubt that this occurs in a considerable proportion of cases

These results indicate that the present methods of dealing with the mastoid cavity after operation are most unsatisfactory

It would seem that, in any mastoid operation, considerable benefit should result if all raw surfaces, whether of bone or soft tissue, could be provided with a covering of *stable* skin at the time of the operation Not only should healing be rapid and complete, but there should be no tendency for the lining to break down at a later stage accompanied by attacks of acute inflammation in the mastoid cavity It also follows that rapid healing in and around the area might well improve the final hearing results by avoiding prolonged infection One of the authors (Beales 1957) has commented on the difference in the behaviour of the skin graft after myringoplasty and after tympanoplasty It was found that the superficial epithelium of the graft desquamated several weeks after tympanoplasty and the graft surface looked like 'raw meat ' This change in the graft did not occur after myringoplasty where there was no raw area left to produce infection of the surface of the graft Since adopting the new technique to be described below, the tympanic grafts have shown little tendency to undergo these changes and hearing has improved more rapidly

A method of obtaining rapid healing by providing all raw surfaces exposed during any form of mastoid surgery with a stable skin cover will now be described The method involves no special training in plastic surgery and adds only about 20 minutes to the normal operating time

Technique—The Post-Auricular Flap Operation

A modified form of hypotensive anæsthesia using the "lytic cocktail" has been employed in these cases and has proved safe and satisfactory. Hypotension though extremely helpful is by no means essential.

The operation as used with tympanoplasty, is carried out as follows:

1. A skin graft of medium thickness and about 10 × 5 cm. in size is first cut from the thin hairless skin of the inner aspect of the upper arm. This graft is wrapped in a piece of gauze wrung out in saline and is placed on one side for later use—a portion will be used as the graft for the tympanic cavity, and the rest will provide skin cover for the secondary defect on the deep surface of the pinna at the end of the operation.

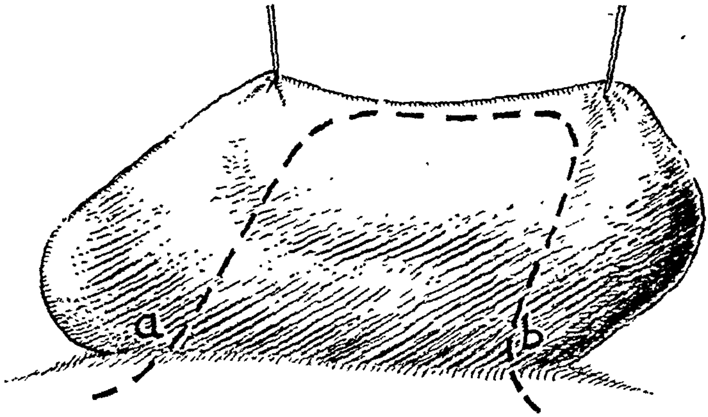

FIG. 1.

The position of the post-auricular flap is marked on the pinna with ink before it is cut. *a–b* shows the line from where a thin strip of skin is denuded of its superficial epithelium in order to close the fistula at the end of the operation.

2. An oblique post-auricular flap, consisting of the skin and subcutaneous tissue of the deep surface of the pinna superficial to the perichondrium, is now raised; it is taken from the whole width of the middle two-fourths of the pinna and its pedicle lies over the mastoid process just behind the retro-auricular groove (Fig. 1). The flap is turned backwards and the mastoid bone is then displayed by a vertical incision through the retro-auricular muscles and the periosteum, which are then separated from the bone by a periosteal elevator. At the anterior border of the mastoid process, the posterior wall of the external auditory canal is exposed and its lining separated from the posterior, superior and inferior walls of the bony portion of this canal by blunt dissection as far as the tympanic ring (Fig. 2).

A meatal flap, based externally, is now cut from the skin of the whole width of the posterior wall of the canal, extending a little on to roof and floor, this flap is made as long as possible, its distal end lying near the tympanic membrane

A mastoid retractor is then inserted, care being taken to prevent pressure on the pedicle of the turned-back post-auricular flap by engaging the posterior jaw of the retractor against the retracted edge of the mastoid periosteum (Fig 3) A second mastoid retractor, placed at right angles to the first, completes the exposure

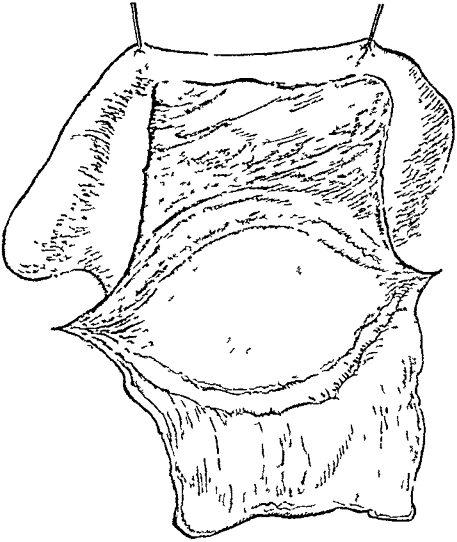

FIG 2

The flap is turned backwards and the mastoid bone is exposed The soft tissue lining of the posterior meatal wall can be seen just before it is dissected anteriorly towards the tympanic membrane

Note the thick periosteal cuff in which the posterior jaw of the mastoid retractor will engage, thus protecting the flap from pressure and damage to its blood supply.

3 The tympanoplasty is now completed in the usual way as described by one of the authors in a previous publication (Beales, 1957) A piece taken from one end of the split skin graft which was cut as the first step

in the operation is used for the reconstruction of the tympanum and is so arranged that its posterior edge extends backwards for a short distance into the adjacent mastoid cavity. The graft is held in place by means of a gelatin foam pack and an outer pressure dressing of ribbon gauze in paraffin, the free end of which is brought out through the external auditory meatus. The mastoid retractors are then removed.

During this part of the operation, the assistant should occasionally raise the turned-back post-auricular flap for a few seconds to straighten out its pedicle and relieve any embarrassment to the blood flow through it; this can be done every 5 or 10 minutes without interfering with the activities of the surgeon. The raw surface of the flap should also be moistened with saline from time to time.

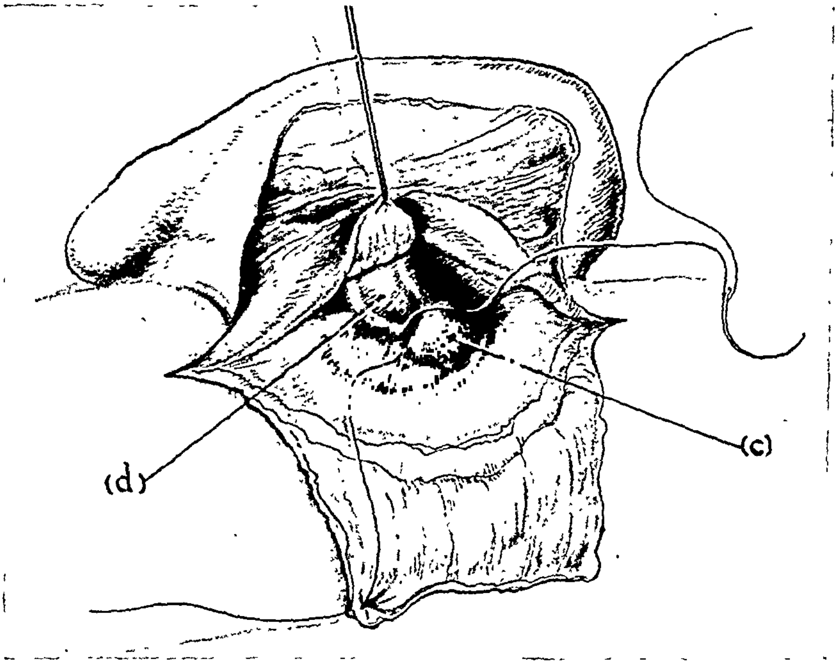

FIG. 3.

The mastoid cavity is seen and is ready to be lined by the flap. (*c*) marks the tympanic cavity with the graft in position extending posteriorly for a short distance. The suture shown passes between the distal border of the flap and the lower cut edge of the lining of the auditory canal (*d*) as near the tympanic membrane as possible.

4. The tympanoplasty having been completed, the raw surfaces (i.e. the mastoid cavity and the soft tissues on the deep surface of the concha) which were created in the process of exposing the tympanic cavity are covered with the post-auricular and meatal flaps which were raised previously. The meatal flap is turned backwards to cover much of

the raw surface on the deep surface of the concha and it is held in that position by a single plain 4/0 catgut suture passed through its apex (Fig. 4). The sharp upper and posterior bony margins of the mastoid cavity are reduced by means of a dental burr. The post-auricular flap is then drawn forwards to cover this saucerized mastoid cavity by means of a plain 4/0 suture passed between the flap and the lower cut edge of that part of the lining, of the external auditory canal which was left *in situ* after raising the long meatal flap early in the operation. This suture, which is carried on a small 5/8 circle needle, when passed between the distal border of the flap and the lower cut edge of the lining of the auditory canal as near the tympanic membrane as possible (Fig. 3), draws the flap deeply into the mastoid cavity until its distal margin lies in contact with the posterior edge of the tympanic skin graft, which as will be recalled, was allowed to extend a short distance posteriorly beyond the tympanum.

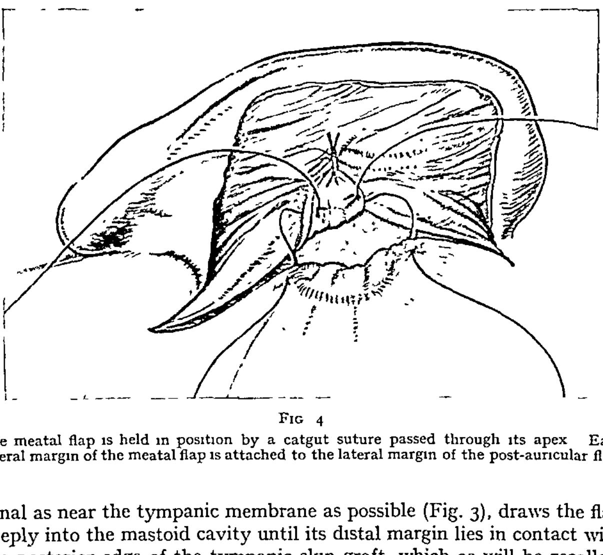

FIG 4

The meatal flap is held in position by a catgut suture passed through its apex Each lateral margin of the meatal flap is attached to the lateral margin of the post-auricular flap.

To complete the skin cover, each lateral margin of the post-auricular flap is curled forwards and attached to the corresponding lateral margin of the meatal flap by a single 4/0 plain catgut suture (Fig. 4).

In this way, all raw surfaces are covered—the tympanic cavity by its skin graft and the large bony and soft tissue defects behind this by a continuous layer of skin provided by the post-auricular and meatal flaps. These flaps are kept in contact with their raw surfaces by a ribbon gauze pack soaked in liquid paraffin; it is inserted gently through the post-auricular wound and the free end then brought out through the external auditory meatus.

5. The operation is now completed except for the fact that (*a*) a fistula, through which the post-auricular flap passes, exists behind the ear between the back of the pinna and the mastoid region and (*b*) there is a secondary defect on the deep surface of the pinna from which the post-auricular flap was raised (Fig. 5).

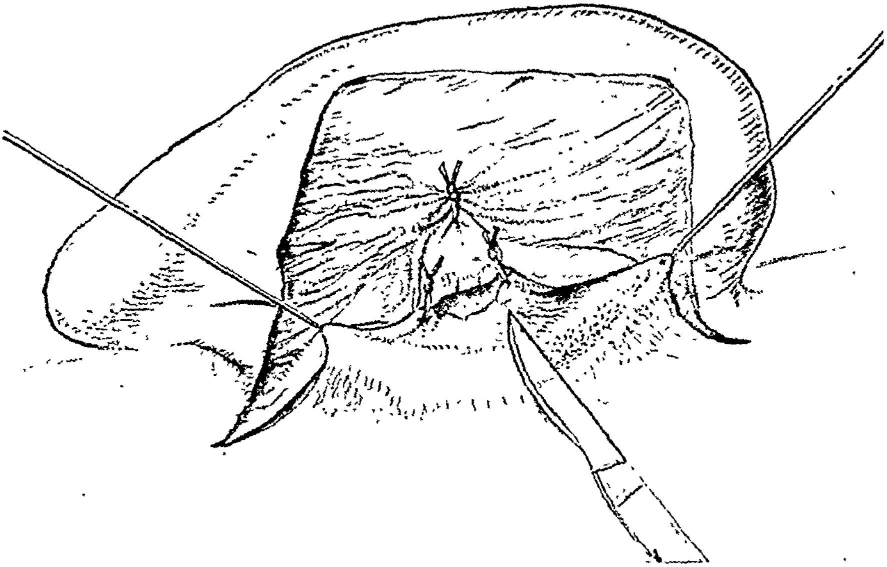

FIG. 5.

A thin layer of epithelium is shaved off the pedicle of the post-auricular flap. This shaved surface is about 0·5 cm. broad and it extends across the whole width of the flap.

The fistula is closed in the following way. The epithelial surface of the pedicle of the post-auricular flap which forms the posterior wall of the fistula is removed by shaving a very thin layer off the surface of the pedicle by hand, using a scalpel with a number 10 blade. Shaving is facilitated by lubricating the surface of the skin with a little vaseline and by putting the pedicle on the stretch in a transverse direction by an assistant gently pulling in opposite directions on two skin hooks, one inserted on each edge of the pedicle (Fig. 5). The shaved surface should be about 0·5 cm. broad and, when the hooks are released, it is seen to be covered with a multitude of points of capillary bleeding. It is important to ensure that small islands of surface epithelium are not left behind. The external (posterior) edge of the shaved strip is then sutured to the raw area on the deep surface of the concha 0·5 cm. behind the apex of the stitched back meatal flap by three plain 4/0 catgut sutures which do not pierce the whole thickness of the pedicle but pass very superficially through its dermal layer only—these superficially placed sutures will not interfere with the blood supply of the post-auricular flap (Fig 6) The two surfaces—the raw deep surface of the concha and the shaved pedicle—fuse rapidly and are firmly healed together at the first dressing

It should be noted that the accurate shaving of the epithelial surface of the pedicle of the post-auricular flap can be a little difficult when done at this stage as the flap is mobile and has to be steadied by hooks as described The operator will probably find it easier to shave the area at the end of step 1 of the operation (see above) before actually raising the post-auricular flap If this method is adopted, the skin graft is first cut from the arm, the surface of the skin of the posterior surface of the pinna immediately anterior to the post-auricular groove is shaved (Fig 1) and the post-auricular flap is then raised

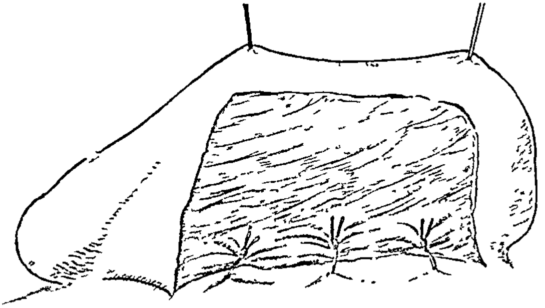

Fig 6

The posterior edge of the shaved strip is sutured to the raw area on the deep surface of the concha just behind the apex of the stitched back meatal flap by three 4/0 plain catgut sutures

6 Finally, the remainder of the skin graft cut from the arm at the beginning of the operation is applied to the secondary defect on the deep surface of the pinna and held in place by a few silk sutures (Fig 7)

7 A pressure dressing is then applied A layer of tulle gras is first placed over the surface of the pinna and is folded round the helix so as to

cover also the skin graft on its deep surface. Pieces of cotton wool, which have been placed in sterile water and then wrung out, are placed firmly behind the pinna to support the post-auricular skin graft; other pieces of the wool are also placed over the external surface of the ear in order to fill the concha and other depressions. The whole area is then covered with a large flat piece of moistened wool and a firm bandage applied.

Post-auricular and meatal flaps can be used in exactly the same way to provide cover for the raw areas created in the fenestration and radical mastoid operations.

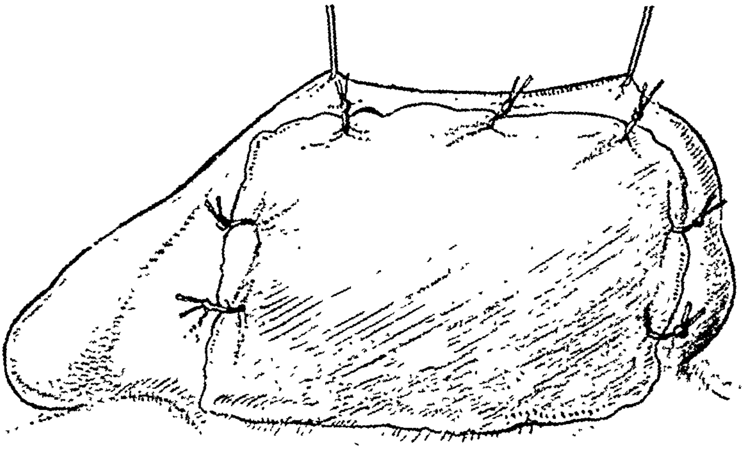

FIG. 7.
The defect on the deep surface of the pinna is covered with a Thiersch graft held in place with a few sutures.

Post-Operative Treatment and Progress

The dressings are removed on the seventh post-operative day. It will then be found that the skin graft on the deep surface of the pinna has "taken" and any redundant portions of the grafts are removed with scissors. The fistula in the post-auricular groove, which was closed by suturing the raw surface on the deep surface of the concha over the shaved pedicle of the post-auricular flap, will be found firmly healed.

Conclusions

A method of providing a skin lining for the mastoid cavity has been described and the writers have treated 25 patients by the technique during the past 6 months. Though the number of patients so far treated is small, the early results have been so satisfactory that the authors have considered it useful to present their impressions of this method of treatment.

The operation obeys the important surgical principle that all exposed raw surfaces should be covered with epithelium and should not be allowed *to heal spontaneously. In addition, the post-auricular flap introduces* a new and copious blood supply to the mastoid region. As a consequence, any early discharge rapidly ceases resulting in a dry ear within a month *in the average case. Quite apart from avoiding the possible effect of* chronic infection and discharge in the mastoid cavity on the final hearing result, there has been a considerable reduction in the number of attendances made by the patients for out-patient dressings.

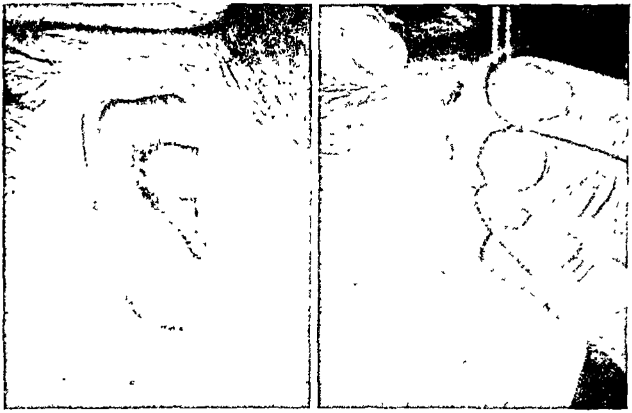

FIG 8 (*a*) and (*b*).

These photographs, taken five months after operation, show a pinna of normal appearance. The fistula is soundly closed and there is minimal scarring of the post-aural region.

Inspection of the mastoid cavity through an aural speculum has shown a lining of normal healthy skin which is quite unlike the injected friable lining seen in so many cases treated on conventional lines. Though the patients have not yet been followed up for a long period, the skin lining the mastoid has appeared so like normal skin that it seems probable that it will remain permanently stable.

After a considerable experience of cases operated on by standard procedures, one of the authors (P.H.B.) feels convinced that the hearing results in patients treated by the post-auricular flap method are better.

The post-auricular flap operation produces a pinna of satisfactory appearance. The external meatus is normal in size and the ear itself does not show a pinned-back appearance, but projects from the side of the head as it did before operation (Figs. 8(a, b) and 9(a, b)).

In 1952 after a discussion between one of the authors (W.H.) and Mr. J. D. Gray a two stage operation using the post-auricular flap to cover the radical mastoid cavity was tried. Two cases were treated but the matter was never pursued.

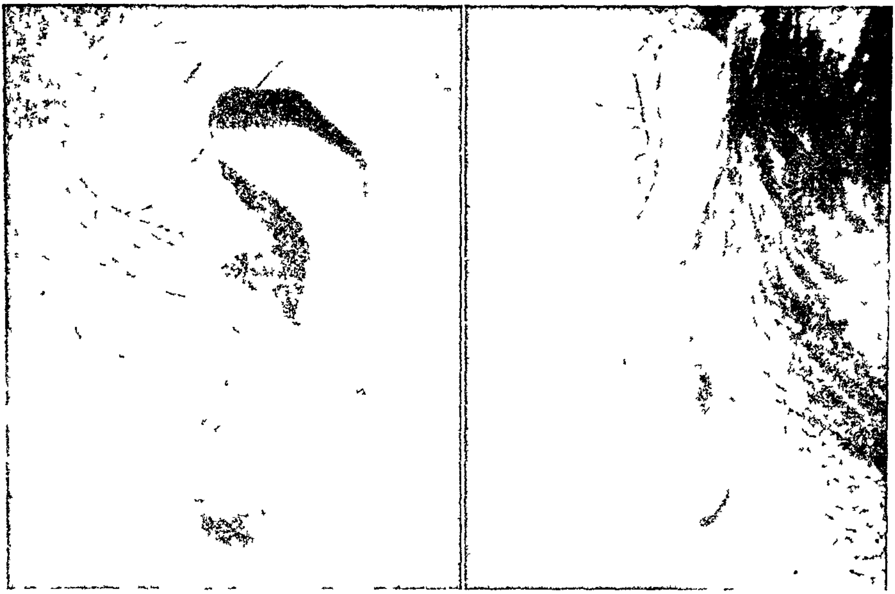

FIG 9 (*a*) and (*b*)

These photographs taken four months after operation, show a pinna of normal appearance which is not pinned back in any way The post-aural region presents a good cosmetic appearance and further improvement will occur with passage of time The mastoid cavity is dry and lined by good quality skin

Following this study of a short series of cases, it is intended to publish long-term results at a later date.

Acknowledgements

The authors wish to thank Mr. F. McGuckin and Mr. I. B. Thorburn for their figures relating to healing after various types of mastoid operation.

We are grateful to Dr. W. H. Baillie, consultant anæsthetist to Doncaster Royal Infirmary, for his help, and we would also like to thank Mr. A. F. Foster, medical artist to the United Sheffield Hospitals and Doncaster Royal Infirmary for his operative drawings.

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SURGICAL EXPOSURE OF THE FACIAL NERVE

By ROBERT C KRATZ, M D, (Newport, Kentucky)
and
STEPHEN HOGG, M D, (Cincinnati, Ohio)*

EXPOSURE of the facial nerve may be divided into three parts: the infraforaminal portion, Tschiassny (1953), distal to the stylomastoid foramen, the vertical portion from the stylomastoid foramen to the

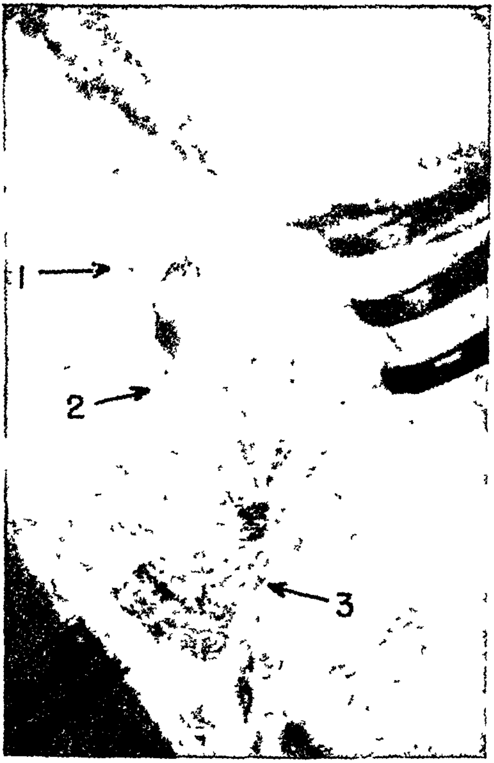

FIG 1

The right ear is retracted upward and anteriorly, showing (1) the tympanomastoid fissure; (2) the facial nerve continuing in the same line as the tympanomastoid fissure; (3) attachments of the parotid fascia to the sternomastoid muscle aditus ad antrum and the horizontal portion deep and anterior to the aditus ad antrum

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* From the Department of Otolaryngology of the University of Cincinnati College of Medicine.

The infraforaminal portion can best be exposed through a post-auricular incision beginning 1 cm posterior and inferior to the superior attachment of the auricle, descending to the angle of the jaw, and anteriorly along the inferior margin of the mandible The first part of the incision is kept anterior to the muscle attachments of the sternocleidomastoid muscle to the mastoid process A blunt elevator is used to identify the tympanomastoid fissure The incision should not enter the parotid gland or the sternomastoid muscle, but follow the natural dissection planes to a point 6 or 8 mm deep to the tympanomastoid fissure At this point the facial nerve will be found coursing in a straight line downward and slightly forward A line drawn from Henle's spine and through the most inferior portion of the tympanomastoid fissure will pass close to the

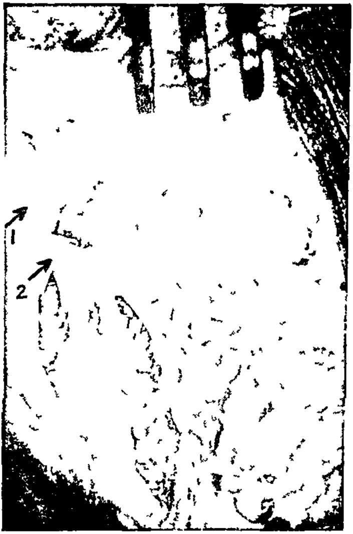

FIG 2

The right ear is retracted upward and traction has been applied to the superficial portion of the gland Note (1) the tympanomastoid fissure (2) the facial nerve continuing in the line of the fissure The five main divisions of the nerve divide in the gland division of the nerve into its temporofacial and cervicofacial divisions (see Fig. 1).

McWhorter (1917) and Bailey (1941) have described the nerve as being "sandwiched" between the deep and superficial lobes of the parotid gland. Treves (1941) and McKenzie (1948) describe it as dividing in the substance of the gland. The important anatomical point is that the superficial portion of the gland can be removed, exposing the nerve without harming it. The skin is separated from the superficial surface of the gland and traction applied to the superficial lobe. Working from the stylomastoid foramen, the septa between the divisions of the nerve are cut, thus exposing the nerve to its periphery and removing the superficial portion of the parotid gland. This manœuvre is illustrated in Fig. 2.

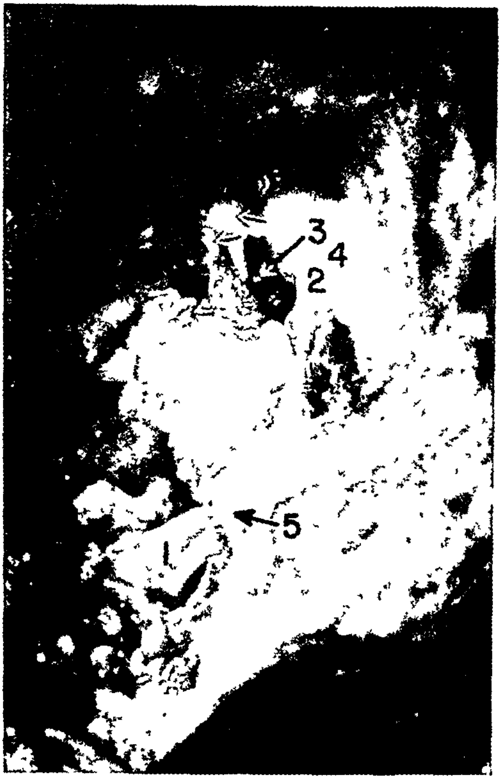

FIG 3

The bone superficial to a line between the inferior portion of the stylomastoid foramen and the lateral semicircular canal has been removed, (2) The facial nerve can be seen beneath the semicircular canal, (3) arrows demonstrate the malleus, incus and stapes, (4) the horizontal semicircular canal, (5) the most inferior portion of the stylomastoid foramen.

Exposure of the vertical portion of the nerve in the temporal bone depends upon the identification of the nerve at the inferior portion of the tympanomastoid fissure and in the aditus ad antrum below the horizontal semicircular canal. The nerve runs nearly in a straight line between these points. On rare occasions, variations from the normal may be seen as described by Kettel (1946). The bone superficial to the two points may be removed with gouge or dental drill. We have found the Hudson burr helpful in exposing the lateral semicircular canal (Goodyear, 1939).

Exposure of the horizontal portion of the nerve to the geniculate ganglion is accomplished by following the nerve across the middle ear superior to the stapes and oval window, anteriorly to the processus cochleriformis above and medially to which the ganglion lies. This last procedure would require removal of the incus, malleus and drum.

Summary

A method for exposure of the facial nerve in the adult, utilizing anatomical skeletal landmarks, is presented.

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CLINICAL RECORDS

SARCOMA OF THE LARYNX

By I. SIMSON HALL (Edinburgh)

SARCOMA of the larynx appears to be a rare condition and it seems appropriate to record a case recently treated, and at the same time to review the information which is available regarding this form of laryngeal disease.

The present case was seen in the ordinary course of clinical work in the Royal Infirmary, Edinburgh, and the radiotherapy was given under the supervision of Professor McWhirter, while the final histological opinions were given by Dr. R. F. Ogilvie, Senior Pathologist to the Edinburgh Royal Infirmary.

FIG I

The details regarding this patient are as follows:

Mr. R.L., aged 46. Occupation: motor driver. First seen on *25.4.57* with a complaint of "laryngitis" for six weeks. There was no dysphagia, and no post-nasal discharge or catarrh.

Examination showed a proliferating tumour involving the whole length of the right vocal cord. Movement of the cord was good. There was no subglottic spread.

Histological examination showed a poorly differentiated growth suggestive of carcinoma

Treatment was immediately commenced by linear accelerator. Treatment was completed on *19 6.57*

On *12.9 57*. The right cord was normal and movement was normal.

On *14 11 57* There was a smooth reddish swelling on the right vocal cord, just anterior to the mid-point.

On *23 1.58*. There was a large mass on the right vocal cord occupying the position of the cord and encroaching considerably on the airway Tracheostomy was considered. The vocal cord was immobile. (Fig. 1) There were no palpable glands.

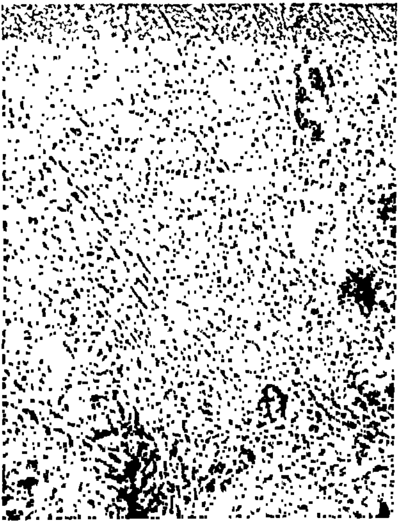

Fig 2

Histological examination showed spindle celled sarcoma. (Fig. *2*)

4.2.58. Laryngectomy with block dissection of glands on the right side was carried out.

Microscopic examination of glands· one gland contained a suspicious deposit.

After some initial difficulty with chest infection, this patient made a good recovery and was discharged from hospital on *9.4.58*.

Comment

As sarcoma of the larynx is not a frequent condition, opinions regarding its clinical characteristics vary widely.

The incidence appears to be under 1 per cent., probably about one half per cent. of all malignant laryngeal tumours.

It should be noted that during the past it has been reported as comparatively common, as e.g. 11 per cent. by Molinie (quoted by Ewing). It is evident that more careful examination in the light of modern pathological opinion suggests that many of these cases were epithelial in origin.

Figi (1933) reports four cases in a series of 713 cases of laryngeal tumour, and Jackson and Jackson (1939) found five cases in their extensive collection. Havens and Parkhill (1941) collected 15 cases reported in English literature.

Types of tumour. Spindle cell sarcoma, osteosarcoma, fibrosarcoma, leiomyosarcoma are reported. According to Ewing, spindle cell sarcoma is limited to the vocal cords and epiglottis.

Clinical features. In general these tumours tend to be smooth and possibly lobulated, and do not ulcerate in the early stages. They ulcerate later and may also commence to grow rapidly. Metastases are late, glands being involved in 15 per cent. of cases (Ewing).

Site. It has been variously reported as occurring in the subglottic region (Negus), interarytenoid region (Imperatori), on the vocal cords (Figi). It may be either intrinsic or extrinsic (Ewing).

Prognosis is probably more favourable than in the case of carcinoma of the larynx.

Treatment. As can be seen by the case reported here, the results of radiation appeared to be excellent, but recurrence was early and rapid.

It is probable that in very localized tumours surgical removal is the most effective treatment, especially as glandular metastases are late.

Examination of the records of reported cases contain several examples of successful local removal, but as the response to radiation is excellent the general principle is that, where voice is of prime importance radiation treatment should be the first treatment tried. It is evident, however, that close watch must be kept upon the patient and surgical treatment employed at the earliest moment where there is suspicion of recurrence.

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A CASE OF GLANDULAR FEVER CAUSING ACUTE RESPIRATORY OBSTRUCTION

By H. M. PARSONS and E. J. A. DODDS (London)

ROBIN S., aged 6, was seen as a routine candidate for tonsillectomy and adenoidectomy, in Out-Patients, in mid-October. He gave a history of recurring throat infections and nasal obstruction, worse at night. His twin came with him and gave a similar history. On examination the two children were mouth breathing due to large masses of adenoids in their post-nasal space and had marked cervical adenitis with fleshy enlargement of their tonsils. They were both put on the waiting list to come into hospital, in due course, for tonsillectomy and adenoidectomy.

Some six weeks later Robin S. was admitted, as an emergency, with a history that he had slept very restlessly overnight and that his breathing was becoming increasingly difficult.

On examination he had a temperature of 104°, a pulse rate of 140, and a respiratory rate of 40. He had no nasal airway at all and gross œdema of all the elements of Waldeyer's ring of lymphatic tissue. The tissues appeared clean with a smooth surface reminiscent of frosted glass. He had marked cervical adenitis, but no other glandular enlargement throughout the body.

An endotracheal tube was inserted with difficulty, but much relief. Some hours later a formal tracheotomy was performed under a general anæsthetic.

Clinically he progressed well after the operation. He was given intramuscular crystalline penicillin and 2·5 mg. of prednisolone morning and evening. He had a nasal airway at the end of the first week. The tracheotomy tube was removed on the seventh day. By that time his liver and spleen had become palpable and glands could be felt in his axillae and left groin. His E.S.R. remained elevated and he was not discharged from hospital for seven weeks. On discharge his tonsils and cervical lymphatic glands were still very much enlarged, but his liver, spleen and axillae had returned to normal. The order of regression of the swelling was, firstly, the lingual tonsils and, secondly, the nasopharyngeal lymphatic tissue. His pharyngeal tonsils persisted in their enlargement and were still enlarged when they were removed on *March 19th*. The actual operation and recovery were normal.

An interesting point is that his twin brother also had glandular fever at the same time, proven with a Paul Bunnell test, but did not have the respiratory obstruction and other findings. He was not admitted to hospital and remained fit throughout. He was submitted to tonsillectomy and adenoidectomy at the same time as his brother.

Pathological Reports

1 His nasal and throat swabs were sterile for pathogens pre operatively

2 Blood counts were as follows

28 11 57	Hæmoglobin, 83 per cent Total W B C ,	22,000 per c mm
	Polymorphs	64 per cent
	Small lymphocytes	10 ,, ,,
	Large lymphocytes	3 ,, ,,
	Monocytes	3 ,, ,,
	Eosinophils	0 ,, ,,
	Basophils	0 ,, ,,
	Atypical lymphocytes	14 ,, ,,
14 12 57	Hæmoglobin 74 per cent Total W B C ,	9 200 per c mm
	Polymorphs	26 per cent
	Small lymphocytes	32 ,, ,,
	Large lymphocytes	16 ,, ,,
	Monocytes	7 ,, ,,
	Eosinophils	0 ,, ,,
	Basophils	0 ,, ,,
	Atypical lymphocytes	19 ,, ,,

3 Paul Bunnell test showed the typical reaction of glandular fever Heterophile antibodies completely absorbed by ox cells, but not by guinea-pig kidney

4 The tonsils on section showed

Robin S In both tonsils the germinal centres are rather larger and more active than usual and the stroma contains large numbers of large lymphocytes with nuclei showing features associated with glandular fever

Roger S The tonsils show features similar to those of his brother, viz those associated with glandular fever

Since 1941 six other cases of glandular fever causing acute respiratory obstruction have been reported in the literature, their ages ranged from 11 months through 2 years to 20 and 22 years—in all cases tracheotomy had to be performed In the earlier cases a diagnosis of laryngeal diphtheria was made prior to the final diagnosis being established by the Paul Bunnell test and blood counts

I should like to take this opportunity of expressing my thanks to Dr Donald Bateman for his help with this case, for his skilful passing of the original endotracheal tube and for the anæsthetics for the tracheotomy and the tonsillectomy and adenoidectomy

I should also like to express my gratitude to the nursing staff involved and to my friends and colleagues, the practitioners of the Waltham Abbey district

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FOREIGN BODY IN MAXILLARY ANTRUM

By C. R. SALKELD (Bournemouth)

ON *January 21st, 1958*, M.G., a male aged 34, was admitted to the Dorset County Hospital, having collided with a bus while driving his own car. Reference to the portrait (Fig. 1) shows the facial injuries. There were cuts and abrasions of the left upper eyelid, the forehead and the nose. Close inspection of the left lower eyelid shows a horizontal wound about ½ in. long, partly hidden by the eyelashes. Mr. Colley, the Ophthalmic Surgeon, reports: "Left pupil widely dilated, lens *in situ*. No gross retinal trauma." The skull was X-rayed to exclude fracture and the report by Dr. Rose, the Radiologist, reads as follows: "There is evidence of a large metal F.B. within the left antrum, being directed downwards and backwards, which has apparently penetrated

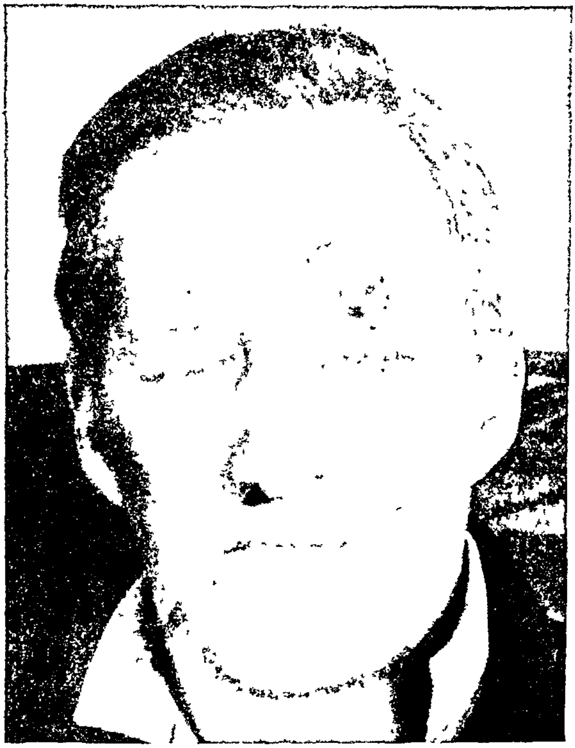

FIG 1